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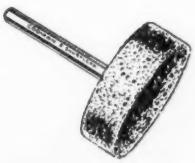

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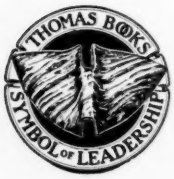

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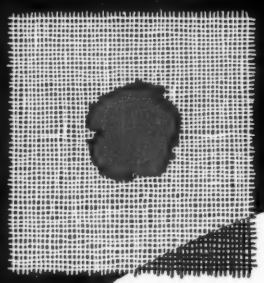

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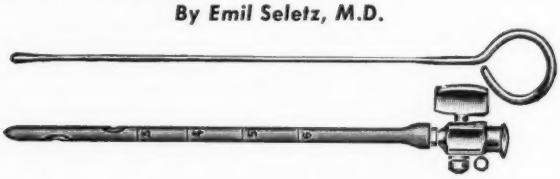

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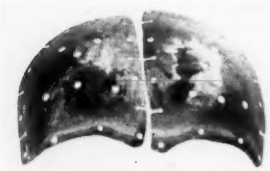

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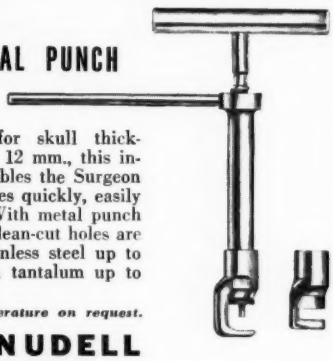

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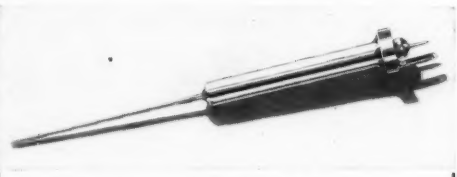

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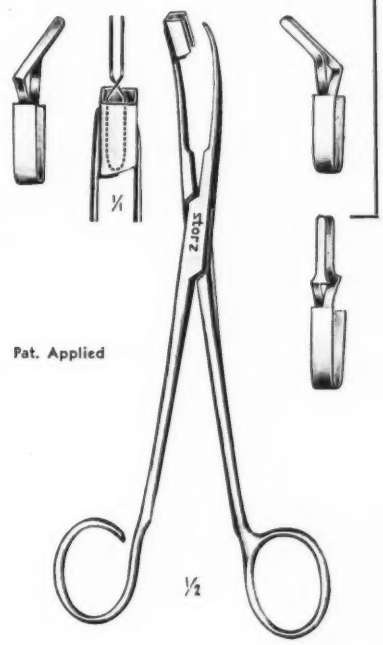

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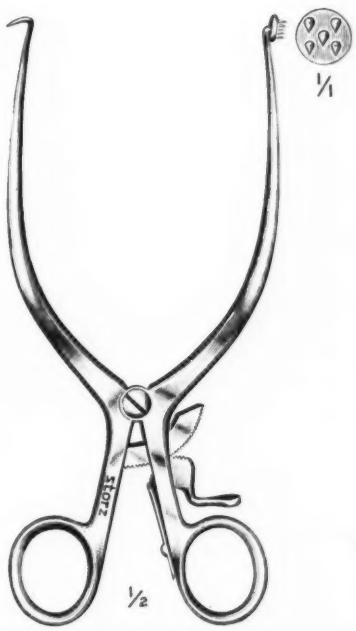

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Professor of Neurology, Columbia University; Director of the Service of Neurology, Neurological Institute, Presbyterian Hospital, New York.

THIS is a book on the present-day application of modern neurologic principles, written expressly for general practitioners and students. Dr. Merritt presents diseases of the nervous system as an integral part of internal medicine, and the entire subject is described clearly in general medical terms. Terminology peculiar to the specialty has been avoided in order that the material might be grasped readily by those not too familiar with the subject.

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An Atlas for Use with the Stereotaxic Instrument

By Jerzy Olszewski, Montreal

93 pages, 57 plates, 5 figures and 4 tables. 1952. Sw. frs. 78.00

From the Introduction:

It is the purpose of this monograph to serve as an anatomical guide for investigators who study the thalamus of the monkey. As most of the work in this field is done with the help of the stereotaxic instrument, special emphasis has been placed on such presentation of the results as will make them most useful to those working with this instrument. The investigator who works with the stereotaxic instrument usually faces two anatomical problems: 1. that of being able to predict accurately during the experiment the site of a lesion or of a stimulus placed in the brain; 2. that of localizing in histological sections the actual site of his intervention, and of comparing his localization with that of other authors.

The solution of the first problem is complicated by a fact, which, in my opinion, has never gained enough attention: that there exists a considerable variation in the proportions of the skull of the monkey, and consequently, data obtained from an experiment on one animal are questionable when applied to another. An attempt to solve this difficulty by the help of X-ray has been made and the results are reported in Chapter III.

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Journal of Comparative Neurology: "Twenty monkey brains were used in the preparation of this atlas. A series of photographs of sections on Heidenhain and Nissl stains are excellently reproduced. The important structures are labeled and the coordinates are indicated at the left and upper margins. The terminology is in Latin based on Walker's nomenclature. Outline drawings are also available for use in stereotaxy. There is no doubt that the material has been carefully prepared and beautifully reproduced. This atlas will be well received by workers in the field of stereotaxy."

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J. Olszewski and H. Meessen: Cytoarchitectonic Atlas of the Rhombencephalon of the Rabbit. 52 p., 15 plates, 69 figures, 1949, Swiss francs 52.00.

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HYDATID CYSTS OF THE BRAIN

ROMÁN ARANA-IÑIGUEZ, M.D., AND JORGE SAN JULIÁN, M.D.
Instituto de Neurología, Facultad de Medicina, Montevideo, Uruguay

(Received for publication December 11, 1954)

EVER since the beginning of this century hydatid (Echinococcus) cerebral cysts have drawn the attention of River Plate authors. In 1901 Morquio[25] published the first case report in Uruguay. Since then, a large number of publications have dealt with the subject.[6,13,23,27,30,31,40] Argentine literature has also often been concerned with brain hydatids from 1893 onwards.[8,11,21,35,36]

Case reports in Uruguay number over 100. On the other hand, Phillips,[34] in Australia, reported 29 cases spread over the last 60 years.

General Notions on Echinococcosis with Special Reference to Cerebral Localization. According to Mackie[22] hydatid disease is most widespread in Algeria, Egypt, Cape Colony, Palestine and Syria, South Australia, Tasmania, New Zealand, Central Europe, Argentina, Uruguay and Paraguay. It is also frequently met with in Spain and Italy. These are all countries noted for the raising of cattle and sheep.

It is interesting to note the distribution of cysticercosis in Latin America. It is very common in Mexico, Peru and Chile, where hydatid disease is a rare occurrence. On the other hand, cysticercosis is hardly ever encountered in Uruguay or Argentina.

Two per cent of all hydatid cysts are found in the brain. The cyst is nearly always solitary in the brain (primary localization); but multiple cysts may develop within the brain if a cyst that is attached to the wall of the left lateral ventricle of the heart ruptures.

According to Dew[15] they are seven times more common in children than in adults. Schroeder and Medoc[40] found 9 adults in 42 cases.

The hydatid cyst grows readily in the brain, a soft, easily displaced tissue; hence, it may attain a large size, keeping its normal hyaline cyst capsule with no daughter vesicles. Nearly every complicated case is the result of rupturing a single cyst.[15] Among our 13 cases we found one suppurating cyst, with daughter vesicles, a very rare occurrence.[20,29]

Infestation with the disease occurs through the ingestion of dogs' feces containing Taenia echinococcus rings. Children may suffer contamination directly from such feces, as do cattle, pigs, and sheep. Contamination of water and foods, principally salads, by such canine feces is probably a more important source of contagion. Contamination by way of the air is still controversial. It could account for pulmonary infection.

The rings deliver eggs, which upon reaching the digestive tract detach from their membrane, freeing a hexacanth embryo 0.025 mm. in diameter. This embryo then passes through the intestinal wall into the blood stream.

After this, the hexacanth embryo has to go through two capillary filters, i.e., the liver and the lung, before it can reach other parts of the body. Hence its high occurrence in liver and lung and rareness in the brain.

In a previous report with Asenjo,[1] the senior author already called attention to the fact that although the hexacanth embryos of the Taenia solium and the Taenia saginata (the latter having the same size as the Taenia echinococcus), penetrate the organism in the same way and have to pass through the same capillary networks, they present very different localizations (nervous system, skin, muscular tissue, eye, but very rarely the liver and lung). It seems logical to conclude that a special histotropism accounts for these differences.

It is impossible to be certain how long it will require for cysts to attain the enormous sizes met with—628 gm. in one of our cases (Fig. 9). Experimentally it has been shown that a cyst takes from 3[14] to 16[28,41] months to grow to 1 cm. in diameter. We suspect, therefore, that cerebral hydatids are at least 3 to 4 years old when they come to the attention of the surgeon.

Hydatid cysts are found most commonly in the parieto-temporo-occipital region. In our 13 cases, 3 involved the frontal lobes, 1 was parietal, and the remainder were temporo-occipito-parietal. In 27 of the 42 cases reported by Schroeder and Medoc,[40] the cysts were found in the temporo-occipito-parietal region. Reggiardo[35] reported an intraventricular cyst but at the Neurological Institute in Montevideo neither Schroeder nor we have had a similar experience. Carrau[9] described a cyst in the sella turcica which he believed to be meningeal. Devé, who examined this specimen later, believed it to be of osseous origin. We shall not deal in this paper with hydatidosis of the cranial bones, which is of rare occurrence.

Microscopically, the hydatid cyst consists of the parasite, a hyaline membrane limited by an adventitia and the germinative membrane, and the reaction of the tissue surrounding the cyst. It should be borne in mind that calcified hydatids have also been observed.[16] The brain is displaced by the cyst, and shows a striking tolerance to it. According to Schroeder and Medoc[40] an area of demyelinization is formed around the cyst, the axis-cylinders remaining intact for a long time. Macrophages and compound granular corpuscles appear then. The vessels and cells assume an arrangement parallel to the surface of the cyst. A moderate fibrillary and cellular hypertrophy of the micro-, oligodendro- and macroglia takes place about the cyst, both close to the cyst wall and at some distance from it. An adventitia surrounds the cyst wall. It can be perfectly isolated during the removal of the cyst. It is made up of a highly vascular connective tissue.

Schroeder and Medoc[40] believe that the changes occurring in the nervous tissue about the cyst are entirely mechanical in origin.

CLINICAL STUDY

The clinical symptomatology attending cerebral hydatid cysts depends on whether we are dealing with a single cyst or multiple ones. We are particu-

larly interested in the single ones, as multiple cysts are rarely found.

Our clinical study includes both children and adults. Of our 13 patients, 10 were children, 15 years of age or younger, and 3 were adults. In one of the latter the cyst was infected.

1. CEREBRAL HYDATID DISEASE IN CHILDREN

The symptomatology usually includes headache, vomiting, lowering of visual acuity, hemiparesis, somnolence and convulsive fits. The objective signs commonly are impairment of general health, somnolence, Macewen's sign, papilloedema, optic atrophy, hemianopsia, hemiparesis, signs of cerebellar dysfunction (tremor and hypotonia), astereognosis and stiffness of the neck.

(a) *Subjective symptoms:*

Headache is observed consistently, and was the earliest symptom in 8 out of 10 cases. Localization becomes difficult because of the age of the patients, the more so when somnolence is present. The area of maximum pain corresponds in some cases to the actual location of the cyst. The headache follows the pattern of headaches caused by intracranial hypertension. It may subside together with the intracranial hypertension, when the latter presents a two-stage evolution as so often happens in children.

Vomiting was present in 9 of our 10 cases. It often is associated with the headaches and improves when the intracranial hypertension subsides.

Lowering of visual acuity. Five children had diminished visual acuity. This symptom may be more frequent but it is sometimes difficult to ascertain in children.

Hemiparesis. In 5 cases the children complained that they were unable to move one half of their body. This disturbance was never found to be intense, not even in one case in which it appeared as the initial symptom.

Somnolence, mental dullness. In 3 instances the parents complained that the children showed mental dullness and were inclined to sleep a great deal.

Convulsions. These were observed in 2 patients. Some writers state that they are rare.[20]

(b) *Objective signs:*

General condition. It was poor in at least 3 of our cases, probably related to the huge size of the cysts. It is usually remarked that cerebral hydatid disease is well tolerated; yet, some writers have observed children whose general health was impaired.

Macewen's sign. This symptom, also called the "cracked-pot" sound, was observed in 3 patients. Sometimes it was found in the area corresponding to the location of the cyst, but in other instances, it was more readily demonstrated in the opposite hemicranium. We also observed one instance of craniotabes.

Papilloedema. It was consistently observed and nearly always was of great intensity.

Optic atrophy. Two patients showed definite optic atrophy.

Hemianopsia. The writers observed homonymous contralateral hemianopsia in half of the patients.

Hemiparesis. In agreement with subjective symptoms, 5 patients exhibited a moderate objective hemiparesis of the pyramidal type. In one case Babinski's sign was elicited bilaterally.

Cerebellar syndrome. This was seen in 5 of the 10 children. In 3 it was associated with hemiparesis. Of these latter, 2 presented both hemiparesis and neocerebellar hemisyndrome on the same side; in the third they were contralateral.

Tremor. This is a striking feature. It is usually bilateral and most clearly seen in the upper limbs. It was observed, with varying intensity, in 6 of the patients.

Hypotonia. In 5 cases it was observed either in two or four limbs.

Astereognosis. This was observed in 2 children.

Stiffness of the neck and Kernig's sign. They were observed clearly in 2 patients. One child showed a palsy of the third cranial nerve, surely caused by a temporal pressure cone; another experienced pains in the cervical region radiating toward the upper limbs. Such spinal pains were also reported by Morquio.[26]

The most important signs observed in our 10 cases may be placed in two groups: those caused by intracranial hypertension, and those of localizing significance.

In children it is not uncommon for the intracranial hypertension to present a two-stage evolution, i.e., after attaining a certain intensity, it improves suddenly and sharply, coinciding with the separation of the cranial sutures. Later the headaches and vomiting return.

Hemianopsia, hemiparesis, astereognosis, and the cerebellar symptoms are of localizing value. The convulsions apparently have only minimal localizing significance. In some cases manifestations that the patient's family refer to as convulsions are in reality generalized, bilateral tremors. These are usually associated with the larger intracranial lesions. Morquio[26] pointed out this possible confusion between tremor and convulsion.

Hypotonia and the cerebellar hemisyndrome were encountered both with occipito-parietal and frontal lesions.

A number of symptoms described in the literature on the subject were not observed by the writers. For instance, we did not observe any psychic disturbances.

We do not believe there are any symptoms typical or diagnostic of cerebral hydatid cysts as some workers do.

The signs of Macewen and the craniotabes, although associated with cerebral hydatid cysts, are a consequence of the great increase of intracranial pressure, so we prefer to deal with them under hypertension symptoms.

2. CEREBRAL HYDATID DISEASE IN ADULTS

We observed 2 adults, 38 and 48 years of age, with cerebral hydatid cysts. The symptomatology corresponded to that of brain tumors. There was

nothing characteristic of this particular condition. In one of these 2 cases there were signs of localized cerebral disease but no papilloedema.

Suppurating hydatid cyst (Fig. 1). In our only case of this type symptoms suggestive of a benign cerebral tumor had been developing for 3 years. The patient's illness began with convulsions. We thought he had a frontal tumor. The only unusual finding was a fever during the 4 days of hospitalization

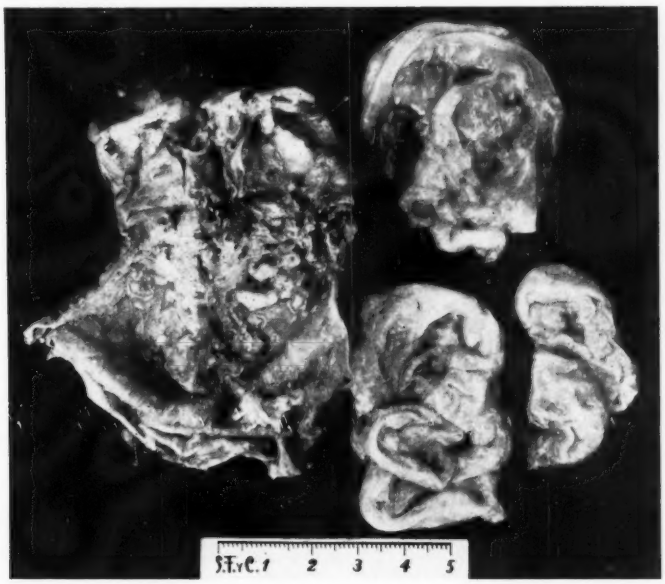

FIG. 1. Specimen of suppurating cerebral hydatid cyst. Notice difference from uninfected cyst in Fig. 9.

prior to operation, coupled with a 10 per cent eosinophilia in the blood. This was unlike Obrador's[29] findings with another suppurating hydatid cyst.

DIAGNOSIS

Morquio[26] came to the conclusion that 50 per cent of cerebral tumors in children are hydatid cysts. During the last two years we have operated upon 18 brain tumors and 8 cerebral hydatid cysts. The clinical characteristics likely to lead to the positive diagnosis of a cerebral hydatid cyst are: 1) symptoms of increased intracranial pressure; 2) diffuse neurological symptoms, not indicative of an exact localization, but revealing a lesion involving a large portion of one cerebral hemisphere or even of both hemispheres; 3) the patients live in rural areas. We have never found a case in Montevideo. 4) The presence of a hyatid cyst elsewhere in the body. According to Dew[15] 80 per cent of cerebral hydatid cysts are associated with cysts in the liver. But of the 13 intracranial cysts reported here only 2 were associated with pulmonary cysts and no hepatic cysts were observed. Furthermore, the

presence of cysts elsewhere in the body is far from conclusive. Schroeder[39] reported the case of a child with a hydatid cyst in the lung and another in the peritoneum. The child had a glioma of the cerebellar vermis.

The general condition of the patient is often only fair. He commonly seeks consultation when the cyst has undergone several years' evolution and attained a large size, giving rise to somnolence, mental dullness and generalized tremor which hinders the correct utilization of the limbs. Of the 3 adult patients, one exhibited obvious mental deterioration. The good general health shown by some patients does not help to differentiate hydatid cyst from other intracranial tumors. Even patients with malignant or metastatic tumors may be in excellent general health.

Pérez Fontana, Rodríguez and Rodríguez Barrios[32] pointed out the occurrence of headache localized at the site of the cyst a few hours after the performance of the Casoni reaction.

DIFFERENTIAL DIAGNOSIS

Since intracranial hypertension is the prevailing manifestation of cerebral hydatid cysts, differential diagnosis is primarily concerned with brain tumors. Tumors of the cerebellum must be given special consideration as they too are more common in children. The erroneous diagnosis of a tumor in the posterior fossa is likely to lead to a serious complication. If ventricular puncture or ventriculography is attempted the hydatid cyst in the parieto-occipito-temporal region is likely to be punctured, scattering the hydatids and giving rise to secondary or daughter cysts. In regions where hydatid disease is a possibility cerebral angiography, not ventriculography, should be used to establish the diagnosis and to differentiate cerebral hydatid cysts from cerebellar tumors. In our 13 cases we generally approached surgical intervention bearing in mind the possibility of cerebral hydatid cysts. Notwithstanding, in one instance we mistook the condition for a posterior fossa tumor and punctured an occipital hydatid cyst while making a ventriculogram.

SUPPLEMENTARY DIAGNOSTIC PROCEDURES

Laboratory Findings. Eosinophilia in the blood, the Casoni reaction and the Weinberg reaction are not of great value. In our 13 cases eosinophilia was looked for in 10 and found in only 3; the Casoni reaction was carried out in 8 and was weakly positive in 1; the only 2 Weinberg tests which were made were negative.

Electroencephalography. According to Fúster, Castells and Gastaut,[18] the typical electroencephalographic findings with a cerebral hydatid cyst are an area of electrical silence surrounded by delta waves. They look upon such a finding as pathognomonic.

Roentgenography. Ordinary roentgenograms may yield highly interesting data, above all as regards the thinning of the vault of the skull, as described by Herrera Vegas,[21] and Phillips.[34] Carrea[10] considers it the stage preceding

the development of parchment-like diploe. In the rare cases of calcified hydatid cysts, regular roentgenography may be very valuable.

Cerebral Angiography. The development of this technique has greatly facilitated the diagnosis of cerebral hydatid cysts. In a previous paper[2,5] we dealt with this subject at length. We concluded that there are several angiographic characteristics of cerebral hydatid cyst (Figs. 2, 3, 4 and 5).

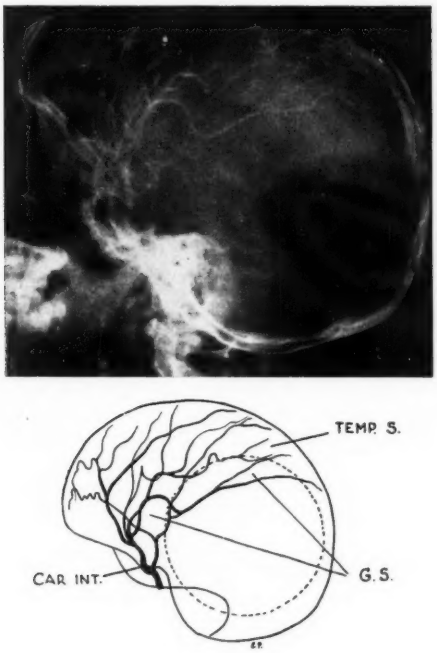

FIG. 2. Angiography showing the enormous displacement of the cerebral vessels and the peculiar aspect of the sylvian group (G.S.).

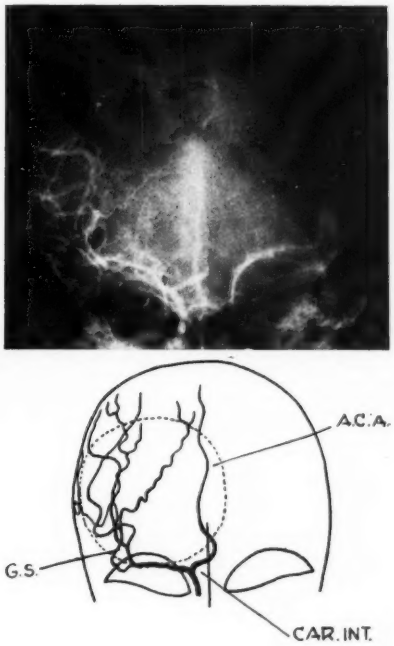

FIG. 3. The vessels of the sylvian group (G.S.) are all crumpled up but even so they keep their peculiarly distended, thick appearance.

1) Complete absence of blood vessels within the lesion.

2) The vessels surrounding the mass are not abnormal as with some intracerebral tumors. They are arranged parallel along the surface of the cyst. Because of the large size of the cyst they are not tortuous and in this respect are similar to those observed in hydrocephalus. The arteries, which Moniz[24] described as "spider legs," have an even diameter throughout their course.

3) There is an enormous displacement of the vessels. Hydatid cysts attain a volume hardly ever exhibited by tumors, although supratentorial tumors in children may attain a huge size, too.[7]

4) The circumferential arrangement of the vessels about the spherical mass is strongly diagnostic of a hydatid cyst.

The above characteristics enabled us to arrive at the diagnosis of hydatid cyst in 6 of our 10 cases. In the 4 remaining cases the technique led us to a well-grounded suspicion of the disease.

Cerebral angiography is a particularly ideal diagnostic method in these cases because it does not entail the risk of puncturing the cyst; as the cysts

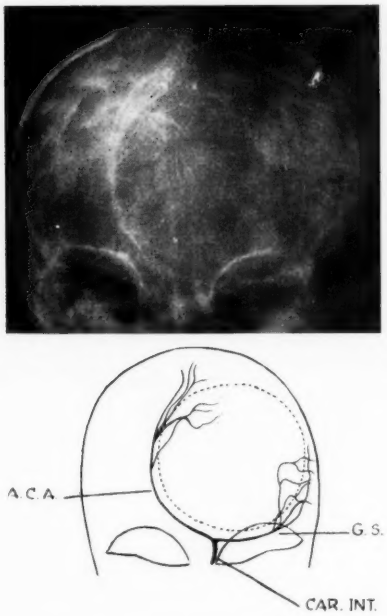

FIG. 4. Circumferential disposition of the anterior cerebral artery (A.C.A.) and the sylvian group.

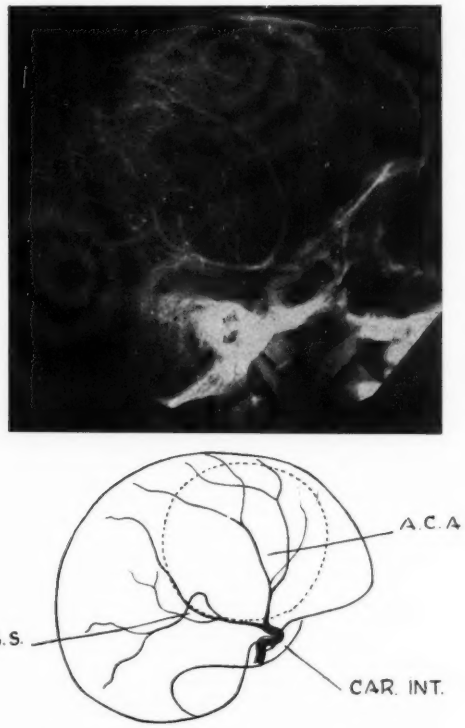

FIG. 5. Lateral view showing the peculiar appearance of the vessels.

are usually supratentorial it is well adapted to these cases; and in most cases it discloses the nature of the condition as well as localizing it.

Although ventriculography is of localizing value and may even disclose some peculiar features as described by Schroeder[39] it is contra-indicated because of the danger of puncturing the cyst.

TREATMENT

The treatment of a cerebral hydatid cyst is surgical. As noted above these cysts are nearly always solitary and when they are removed without ruptur-

ing the wall the patient generally recovers and is cured of his cerebral disease.

In Uruguay, the Dowling[17] or Schroeder[38] techniques were applied until a few years ago. In our opinion, the former is the more advantageous because it enables one to remove the cyst intact, through the technique described in detail by Viale,[42] a disciple of Dowling. A large bone flap is reflected, the dura mater is opened, and then cerebral cortical incisions, that Dowling graphically termed episiotomies, are made. Finally, through gentle maneuvers, placing cotton pledgets between the brain and the cyst, the latter is induced to come out spontaneously. We have removed two cysts in this ingenious manner, which fulfills all the conditions of ideal treatment.

Prof. Schroeder[38] worked out a technique by which the cyst is punctured after thoroughly walling-off the exposed surface. One per cent formalin is then injected into the cyst and left 5 minutes in the cavity. Then the fluid is evacuated and the cystic membrane is easily removed with the aspirator, to which it adheres.

We applied this technique in 3 cases in which the true condition was not recognized early. It is a useful procedure enabling the surgeon to remove the membrane expeditiously whenever the cyst is ruptured during an operation. However, Schroeder himself admitted the hazards of contamination resulting from the puncture of the cyst.

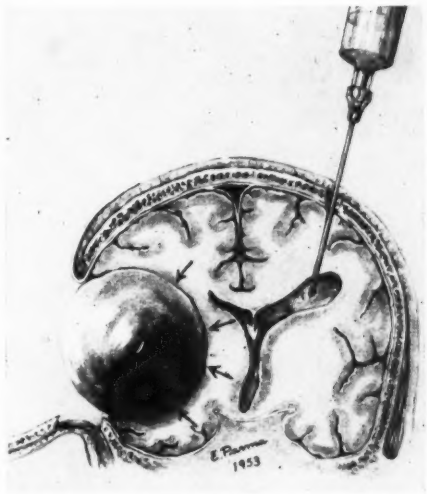

FIG. 6. Diagram of technique proposed by Arana, Rodríguez Barrios and San Julián.[3,4] The cyst is expelled by air injected into the opposite ventricle.

In 1 of the 3 cases in which we used this method, we had suspected a tumor of the posterior fossa and had punctured the cyst during ventriculography. A recurrence resulted which required a second operation 1 year later. Because of this, we consider the techniques reported by da Gama Imaginario,[19] Dew,[16] and Philipov[33] as ill advised.

We propose two techniques for removal of the cyst.[3,4,37]

1. The technique we proposed in 1951 together with Rodríguez Barrios[3,4] (Fig. 6) may be summed up as follows: 1) diagnosis and localization of the cyst by cerebral angiography;[2,7] 2) a very broad osteoplastic flap; 3) contralateral ventricular puncture; 4) exposure of the cyst by radiating incisions of the cortex; 5) placing of the head with the cyst dependent so that gravity will aid in removing the cyst; 6) a gentle injection of air into the ventricle of the opposite cerebral hemisphere; 7) gentle irrigation of saline between the cyst and the cerebral substance.

2. Observation of the above technique convinced us that the injection of saline solution between the cyst and the brain could not only *facilitate* the removal of the cyst but *provoke* it. Accordingly we have managed our 4 most recent cases as follows, with highly satisfactory results[37] (Figs. 7, 8 and 9).

This technique is comprised of the following stages:

1) Localizing diagnosis by cerebral angiography; 2) a very broad osteo-

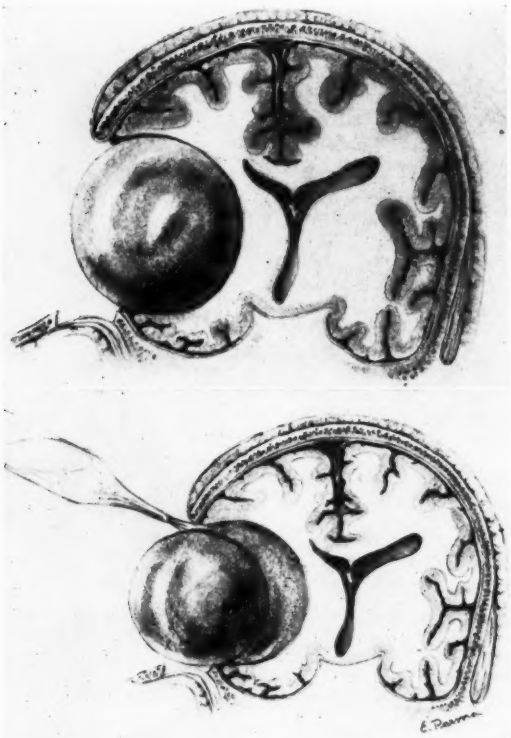

FIG. 7. Diagram of technique proposed by San Julián and Arana.[37] Saline solution is injected between the cyst and the brain.

plastic flap; 3) incisions in the brain substance, only when necessary, and taking care not to make them too extensive but attempting to keep the cavity between the cyst and the brain relatively closed so that the injected saline may collect within this cavity beneath the cyst; 4) placing of the patient with the cyst dependent so as to derive the greatest benefits from gravity; 5) irrigation of saline between the cyst and the brain substance (Fig. 8). Even without the help of gravity the injection of saline solution is highly effective, undoubtedly more so than the injection of air into the

opposite ventricle. Furthermore this technique does not require a contralateral opening for ventricular puncture.

While the previous technique presented the advantage of avoiding any contact with the cyst membrane during surgical maneuvers, in this second method only the saline solution gets in touch with it, thus ensuring the delicacy of the maneuvers, for there is no contact with solid instruments or cotton pledgets. Since the liquid is injected between the cyst and the nervous substance surrounding it, the slight hypertension that develops is less likely to increase intracranial pressure than air injected into the opposite ventricle. Although the number of cases is still low, this latter technique apparently results in a more benign postoperative course than that following the techniques of Dowling or Schroeder, or our first procedure. In the case of very deep cysts, the first procedure may still prove very useful.

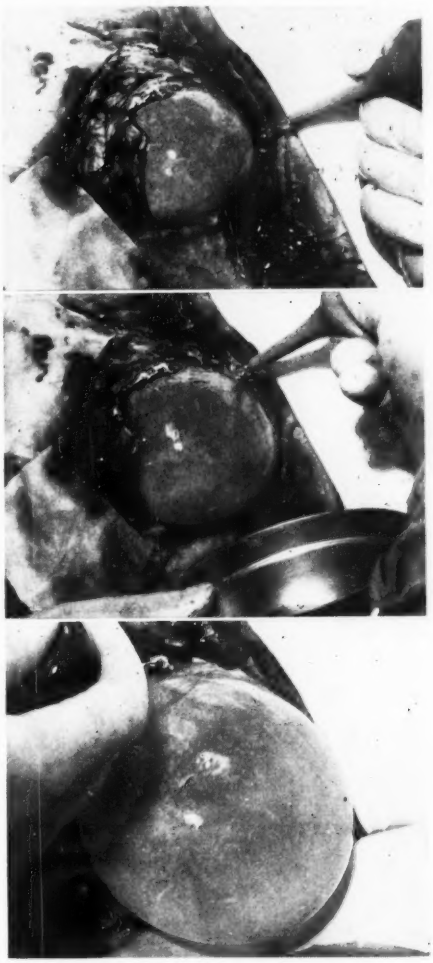

FIG. 8. Illustration of technique devised by San Julián and Arana.[37]

Before closing this paper we cannot help recalling the following remarks, uttered by the Argentinian, Cranwell[12] in 1908.

Hydatid cysts are usually operated upon after attaining such a huge size that the brain has been irreparably damaged. Operation in such cases sometimes only hastens the patient's death. Even those patients who survive are often not actually cured. In fact, in many cases, only the immediate danger is averted and somber complications still await them. Hence, with the long term view we conclude that cerebral hydatid disease is a particularly serious, nearly always fatal condition.

Fifty years later, thanks to the cerebral angiography of Moniz[24] and the present-day neurosurgical technique initiated by Harvey Cushing, and the pioneer work of Dowling in Argentina and Schroeder in Uruguay, we may

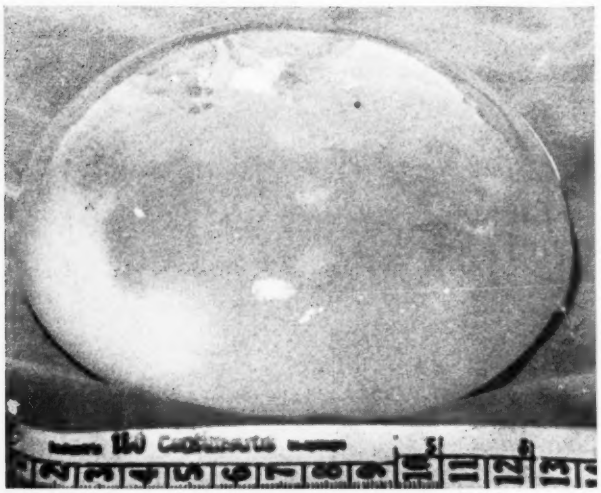

FIG. 9. Hydatid cyst of 628 grams.

state that the dismal outlook of Cranwell has given way to the hope of success in every single case.

SUMMARY

On the basis of 13 observed cases of cerebral hydatid cyst, we have discussed their clinical characteristics and emphasized the importance of angiography for the diagnosis of the disease. Current operative methods have been discussed, and we have proposed two techniques involving: 1) injection of air into the contralateral ventricle; and 2) injection of saline solution between brain tissue and the cyst. We favor this latter procedure because of its simplicity and effectiveness.

The above 13 patients were operated upon and all are living at present.

In one case the cyst was punctured during ventriculography when we suspected a tumor in the posterior fossa. This patient was re-operated upon one year later. There were three cysts in the bed of the primary cyst.

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HEMIPLEGIA CAUSED BY CEREBROVASCULAR THROMBOSIS

AN ARTERIOGRAPHIC STUDY

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With the development of carotid arteriography, the vascular bed supplied by the internal carotid artery has become readily accessible for diagnostic study. As a result, cerebral angiography has provided a rational basis for advances in the therapy of many intracranial lesions. The greatest contribution has been to the management of the primary vascular abnormalities such as aneurysm and the congenital vascular malformations. However, cerebrovascular thrombosis, another clinically important lesion accessible to arteriography, has not been systematically investigated.

A majority of reports in the literature pertaining to thrombosis affecting the cerebral circulation are concerned with occlusions of the internal carotid artery proximal to the cerebral vessels.[7,9,13,16,17,20–22] In 1950 Almeida Lima,[2] in his monograph *Cerebral Angiography*, presented arteriograms illustrating thrombosis of the internal carotid artery, and described the principal characteristics of the clinical syndromes associated with this lesion. In 1951, in a very comprehensive review of the literature, Johnson and Walker[9] found 101 reported cases of internal carotid thrombosis in which the sites of the occlusion were either within a few centimeters of the carotid bifurcation in the neck or within the internal carotid siphon intracranially. While internal carotid thrombosis is undoubtedly more frequent than previously suspected, it is probable that the hemiplegic syndrome clinically considered secondary to thrombosis results more commonly from occlusion of the cerebral portion of the circulation. Although Almeida Lima presented the arteriograms of 3 cases of suspected brain tumor in which occlusion of the intracerebral vessels was found, he did not study cases in which cerebrovascular thrombosis was a primary diagnosis. Wechsler and Gross[23] in 1949 presented 4 cases of "cerebrovascular accident" in which occlusion of the internal carotid or cerebral vessels was demonstrated. In 1951, in an analysis of electroencephalographic changes secondary to cerebrovascular thrombosis, Jones and Bagchi[10] presented clinical data on 10 cases in which thrombosis of major cerebral vessels was proven by arteriography. A similar study was reported in 1952 by Bergouignan *et al.*,[3] and in 1954 more extensive arteriographic

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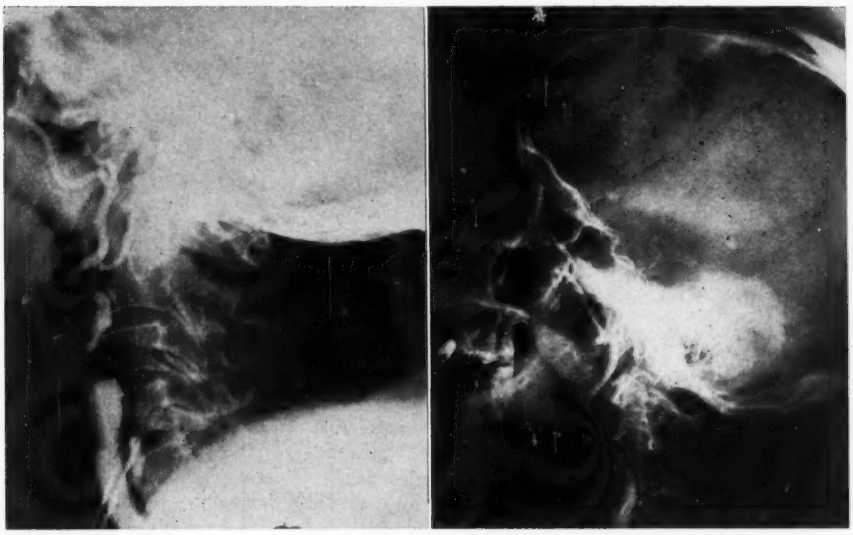


FIGS. 1 and 2. (*Left*) Case 4, Table 1. Complete obstruction of internal carotid artery at common carotid bifurcation. Additional views showed no evidence of retrograde filling of cerebral vessels from the ophthalmic artery on the involved side, but cross-filling into both middle and anterior cerebral arteries from the opposite carotid. (*Right*) Case 6, Table 1. Thrombosis of internal carotid artery in the neck with partial recanalization.

FIG. 3. Case 8, Table 1. Complete obstruction of internal carotid artery in the third portion of the carotid siphon.

studies of carotid and cerebral artery occlusion were reported by Decker and Holzer[5] in the German literature. We believe that more detailed arteriographic analysis of the clinical syndromes resulting from cerebrovascular thrombosis will be of value and present the following data on the vascular anatomy of the hemiplegic "stroke" syndrome.

In a series of more than 500 cerebral arteriograms which we have done during the past 5 years, we have studied 30 patients with hemiplegic

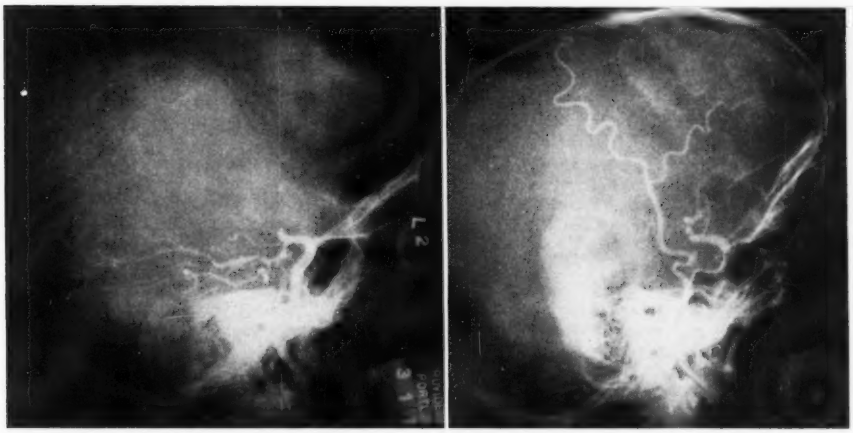
TABLE 1

Abnormal arteriograms showing arterial occlusion proximal to the cerebral circulation

Case	Age Sex	Hemiplegia			B.P. at Time of Study	Arteriograms
		Duration	Mode of Onset	Residual		
1. L.S.	59 F	6 mos.	Gradual Progressive	Complete paresis Severe aphasia	210/120	Bilateral—Right: normal. Left: occlusion int. carotid 1 cm. distal to bifurcation.
2. E.G.	67 M	4 yrs.	Sudden Severe	Slight paresis Severe aphasia	140/70	Bilateral—Right: normal Left: occlusion int. carotid at bifurcation.
3. C.K.	57 M	2 days	Progressive (hours) Severe	Severe paresis Severe aphasia	120/68	Bilateral—Right: normal. Left: occlusion int. carotid 2 cm. distal to bifurcation.
4. V.S.	47 M	25 yrs.	Sudden Complete	Severe paresis Severe aphasia	126/80	Bilateral—Right: normal. Left: occlusion int. carotid at bifurcation.
5. P.E.	47 M	2 days	Progressive Complete	Severe paresis Severe aphasia	142/94	Bilateral—Left: normal. Right: occlusion int. carotid 2 cm. distal to bifurcation.
6. S.N.	49 F	1½ yrs.	Progressive Severe	Severe paresis	148/96	Right: occlusion int. carotid —partial recanalization.
7. F.J.	47 M	2 days	Progressive Moderate	Slight paresis Slight aphasia	136/80	Left: occlusion int. carotid —partial recanalization.
8. M.M.	36 F	1 day	Sudden Severe	Severe paresis	112/60	Right: occlusion in distal portion of carotid siphon.

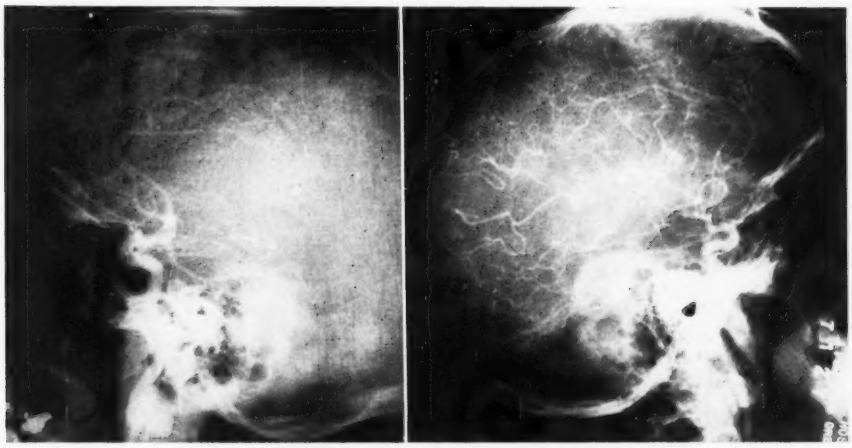
"stroke," all having histories and clinical findings entirely typical of a cerebrovascular thrombosis involving the cortical motor system. Our purpose was first to identify the occluding lesion if possible, and secondly to attempt to correlate the arteriographic evidence with the clinical syndrome.

In the recent literature there are numerous references[6,9,20] to the increased hazards of arteriography in elderly patients, and in the presence of known cerebral vascular disease or occlusion. We have observed certain precautions in this regard, and have attempted to make some evaluation of



FIGS. 4 and 5. (*Left*) Case 9, Table 2. Complete obstruction of internal carotid artery just distal to posterior communicating artery. The posterior cerebral circulation is well shown. (*Right*) Case 11, Table 2. Complete obstruction of middle cerebral artery with markedly reduced and delayed filling of anterior cerebral artery.

the effect of the arteriographic procedure on the clinical status of these patients. The study includes patients from 23 to 85 years of age with one-third being over 60. We have avoided studying patients showing obvious clinical improvement, selecting for arteriography cases in which the neurologic picture had stabilized, or in some instances was deteriorating. Patients



FIGS. 6 and 7. (*Left*) Case 10, Table 2. Complete obstruction of middle cerebral artery. The anterior and posterior cerebral arteries are well filled. (*Right*) Case 18, Table 2. Absence of filling in middle sylvian area. Stereoscopically an area of avascularity, approximately 4×3×3 cm. in size, is clearly demonstrated. The few vessels visualized within this area appear to be filled in a retrograde fashion.

TABLE 2

Abnormal arteriograms showing intracerebral arterial occlusion

Case	Age Sex	Hemiplegia			B.P. at Time of Study	Arteriograms
		Duration	Mode of Onset	Residual		
9. L.J.W.	63 M	1 day	Progressive Complete	Severe paresis Severe aphasia	120/80	Bilateral—Right: normal. Left: occlusion mid. and ant. cerebral (post. cerebral fills).
10. R.E.	67 M	1 yr.	Progressive Severe	Severe paresis Severe aphasia	128/80	Bilateral—Right: normal Left: occlusion mid. cerebral (ant. and post. cerebral fill).
11. H.P.	70 M	1 day	Sudden Severe	Severe paresis Severe aphasia	160/80	Bilateral—Right: normal Left: occlusion mid. cerebral (ant. cerebral—slight filling).
12. C.McI.	46 F	7 days	Sudden Severe	Complete paresis Complete aphasia	160/90	Bilateral—Right: normal. Left: occlusion mid. cerebral (ant. cerebral—slight filling).
13. A.H.	46 M	5 days	Sudden Severe	Severe paresis Severe aphasia	210/130	Bilateral—Right: normal. Left: occlusion mid. cerebral (ant. cerebral—slight filling).
14. E.L.	49 F	11 mos.	Sudden Severe	Severe paresis	150/85	Right: absence first portion of mid. cerebral—poor distal filling (ant. cervical slight filling).
15. P.B.	60 M	2 wks.	Sudden Moderate	Moderate paresis	148/82	Bilateral—Left: normal. Right: reduced mid. cerebral—absent ascending frontoparietal.
16. M.F.	85 M	2 days	Sudden Complete	Complete paresis	128/54	Right: reduced mid. cerebral—no filling of ant. cerebral.
17. F.T.	63 M	1st: 5 yrs.	Sudden Severe	"Recovered"		
		2nd: 3 yrs.	Sudden Severe	Severe paresis Severe aphasia	156/94	Left: reduced mid. cerebral —absent ascend. frontoparietal.
18. F.S.	41 M	12 days	Sudden Complete	Complete paresis Complete aphasia	138/90	Bilateral—Right: normal. Left: reduced mid. cerebral —defect in Sylvian group

were studied at intervals ranging from 24 hours to 25 years following the onset of hemiplegia. In the entire group we found no evidence that the arteriography had a deleterious effect on the patient, and in fact were sur-

TABLE 3

*Normal arteriograms**

Case	Age Sex	Hemiplegia			B.P. at Time of Study	Arteriograms
		Duration	Mode of Onset	Residual		
19. F.H.	63 M	6 yrs.	Sudden Complete	Severe paresis	180/120	Right: normal pattern. Caliber irregularity—filling defects in lumen of siphon.
20. V.S.	47 F	2 wks.	Sudden Complete	Severe paresis Severe aphasia	160/100	Left: normal pattern. Vessels very small caliber *distal* to siphon.
21. C.E.S.	35 M	1st: 4 mos.	Sudden Moderate	"Recovered" in 2 days		
		2nd: 2 days	Sudden Complete	Severe	110/70	Left: normal.
22. M.S.	70 F	2 yrs.	Sudden Complete	Moderate paresis Severe aphasia	210/110	Left: normal.
23. A.O.	38 M	2 yrs.	Sudden Severe	Mild paresis Severe aphasia	130/70	Left: normal.
24. V.D.	50 F	3 wks.	Sudden Complete	Severe paresis	115/78	Right: normal.
25. M.H.B.	32 F	10 days	Sudden Severe	Moderate paresis	116/68	Bilateral: normal.
26. G.S.	65 M	5 mos.	Gradual Moderate	Moderate paresis Moderate aphasia	170/100	Bilateral: normal.
27. W.S.	31 M	5 wks.	Sudden Complete	Moderate	120/80	Bilateral: normal
28. I.G.	46 F	18 mos.	Sudden Complete	Mild paresis Severe aphasia	142/84 (previous known hypertension)	Bilateral: normal.
29. S.S.	67 M	1 yr.	Sudden Complete	Severe paresis	124/80	Bilateral: normal.
30. W.L.	31 M	4 wks.	Sudden Severe	Moderate paresis	130/80	Bilateral: normal.

* Cases 19 and 20 show changes which may be significant in relation to the hemiplegia, although specific arterial occlusion is not demonstrable.

prised in approximately one third of the cases to note significant improvement in the clinical and neurologic status of the patient appearing directly following the procedure.

Arteriography was done percutaneously in all cases, using 35 per cent diodrast with serial films and in almost all instances with stereoscopic views. Bilateral arteriograms were not done in the earlier cases and in 12 patients only the side of pathology was investigated. In the remaining 18 patients bilateral studies have been made usually in a single procedure. Bilateral arteriography in a single procedure aids significantly in evaluation of the lesion, and in our experience has presented no additional hazard to the patient.

Eighteen of the 30 patients studied showed well-defined vascular obstruction. In 8 cases the thrombosis was "extracerebral" (Table 1): 7 of these lesions involved the internal carotid artery at or distal to the bifurcation of the common carotid in the neck, and 1 involved the distal portion of the carotid siphon intracranially. Typical lesions in this group are illustrated in Figs. 1, 2 and 3. In half of these patients the hemiplegia was intermittent and progressive in onset, sometimes over a period of hours, but more often over a period of days, weeks, or months. In the group of patients with "intracerebral" obstruction (Table 2) and in those with "normal" arteriograms (Table 3) such a mode of onset was much less common.

In 10 cases the obstruction was distal to the siphon and these are summarized in Table 2 as "intracerebral" thrombosis. Typical lesions from this group are illustrated in Figs. 4, 5, 6, and 7.

The remaining 12 patients had arteriograms which were interpreted as being "within normal limits" (Table 3), although 2 of them showed changes that we believe may have pathological significance in relation to the hemiplegia.

DISCUSSION

Failure to demonstrate vascular occlusion in such a large proportion (40 per cent) of patients with well-defined hemiplegic "stroke" may be attributable to our inability to visualize the finer vascular bed, or to recognize subtle but critical changes in vascular pattern. However, the appearance of a "normal" vascular pattern in spite of marked loss of function, is not inconsistent with experimental observations that transient occlusion of the middle cerebral artery frequently results in persisting hemiparesis, even though blood flow subsequently appears fully re-established.[8]

The importance of factors other than the occlusion per se in determining neurological deficit is emphasized by the clinical experience of Dandy, Poppen and others with the influence of blood pressure levels on tolerance for surgical ligation of the anterior cerebral arteries.[4,12]

This series is too small to permit reliable evaluation of many factors that may relate to the severity and course of the clinical disability. However, one difference between the "normal" and abnormal arteriogram groups is sug-

gested by our data. Although the hemiplegia initially seems equally severe, the potential for recovery appears greater in patients with "normal" arteriograms than in those in whom major vascular occlusion is demonstrable. More than half the patients in the "normal" group had shown significant reduction in disability at the time of study while only 2 of the 18 patients in the "abnormal" group had shown comparable improvement.

The presence or absence of arterial hypertension might be expected to have prognostic significance in relation to the severity and course of hemiplegia following thrombosis. Arterial hypertension (150/90 or above) was present in nearly half of the patients at the time of study, and may have been present prior to the "stroke" in others. However, in this small group the hypertensive patients did not have a higher incidence of gross vascular occlusion, nor was the hemiplegia necessarily of greater severity in the hypertensive than in the normotensive patients.

The errors in clinical diagnosis that we have made in the course of this study fall into two groups. First, patients in whom no occlusion is demonstrable on carotid arteriograms, may later prove to have occlusion in the vertebral-basilar system. More critical neurological examination and follow-up will establish the correct diagnosis. Secondly, a patient in whom definite occlusion is demonstrated may subsequently prove to have an infiltrating tumor of the hemisphere. The onset of the hemiplegia may be entirely consistent with thrombosis, and there may be no significant displacement of major vessels at the time of study. More critical review of these arteriograms, however, has shown irregularities in vascular pattern in addition to the occlusion, and in retrospect we believe that such errors in diagnosis should be avoidable.

Investigations of many factors that relate to the mechanisms of thrombosis and vascular degeneration suggest methods of approach to future therapeutic attack on this problem. Experimental studies of intravascular aggregation and adhesiveness[19] resulting in an increased blood viscosity[18] of cerebral blood flow and gaseous exchange,[11,14,15] and of vascular sclerosis[1] are among those that indicate that the incidence of vascular occlusion and the secondary damage which follows might be reduced.

Our experience leads us to propose that further clinical study of cerebrovascular thrombosis be carried out—first to enlarge our knowledge of the vascular changes associated with the "stroke" syndrome, and ultimately to apply available data in an effort to reduce resulting disabilities.

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INTRACRANIAL COLLECTIONS OF FLUID WITH LOCAL BULGING OF THE SKULL

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LOCAL bulging of the skull is an uncommon manifestation of an intracranial space-occupying process. It is commonest in children and the usual causes are chronic subdural hematomas and cerebral tumors.[6,8] The skull changes in cases of juvenile chronic subdural hematomas have been described by Davidoff and Dyke[6] and Bull.[1] Hardman[7] reported a case in which similar changes in the skull were associated with what was believed to have been a chronic subdural hygroma—a closely related condition. Like cases have been recorded,[1,2,3,10,11] and they will be reviewed later. We have had 2 such instances and from a study of them fresh suggestions as to their nature will be made.

CASE REPORTS

Case A (NS 5234). A boy aged 8 years was admitted on July 14, 1953 with a 3 months' history of swelling of the head above the left ear. It was thought that this had increased in size. There were rare headaches. He had no other complaints and he was in the normal class for his age.

Examination. The sole physical sign was a painless swelling above the left ear. Roentgenograms of the skull showed thin bulging bone in the left temporal region above and in front of the left ear (Fig. 1). The middle fossa was not expanded forwards. Left carotid arteriogram showed that the middle cerebral artery was elevated from its origin and there was a slight shift of the anterior cerebral artery to the right (Fig. 1). No tumour circulation was seen. EEG showed excessive slow activity in the left temporoparietal leads. The pre-operative diagnosis was of a temporal glioma.

Operation. A temporal bone flap was raised. The bone was thin and bulged outwards. A normal-looking dura mater was opened. At the site of the temporal pole was a bluish transparent cyst. The arachnoid swept off the temporal and frontal lobes to make up the outer wall of the cyst (Fig. 2). There were some small vessels running in the wall to the middle cerebral vessels. The cyst was opened and contained clear fluid. The medial part of the cyst communicated with the subarachnoid space over the free edge of the tentorium. Cerebrospinal fluid welled up into the cyst during the operation. The anterior part of the temporal lobe was absent and such lobe as was present had a normal gyral pattern. There did not appear to be any arachnoid over the brain that walled the cyst. Beside the tentorium, the olfactory, optic and oculomotor nerves and the carotid artery and its bifurcation were visible without retracting the brain. The arachnoid wall was stripped away. In closing the wound, the bone flap was wired into place.

The *postoperative course* was uneventful and he was well a year later.

FIG. 1. *Case A.* Left carotid arteriogram (anteroposterior view) showing elevated course of middle cerebral artery and slight shift of anterior cerebral artery to right. The bulging of the left temporal area can be seen.

Case B (NS 6522). A boy aged 14 years was admitted on April 9, 1954 with a story that at the age of 4 years he had been hit on the head with a toy pistol. Since then his parents had noticed that the left temple had been swollen. There had been a dubious increase in size. There were no other complaints and he was in the normal class for his age.

FIG. 2. *Case A.* View of operation showing the cyst at the temporal pole. This is covered with arachnoid.

FIG. 3. *Case B.* Left carotid arteriogram (anteroposterior view) showing elevated course of middle cerebral artery and the space between brain and skull. Some bulging of the skull can be seen in the left temporal area.

Examination. There was considerable protrusion of the head in the left temporoparietal region. There were no other physical signs. Roentgenograms of the skull showed marked bulging of the left temporoparietal area with thinning of the bone

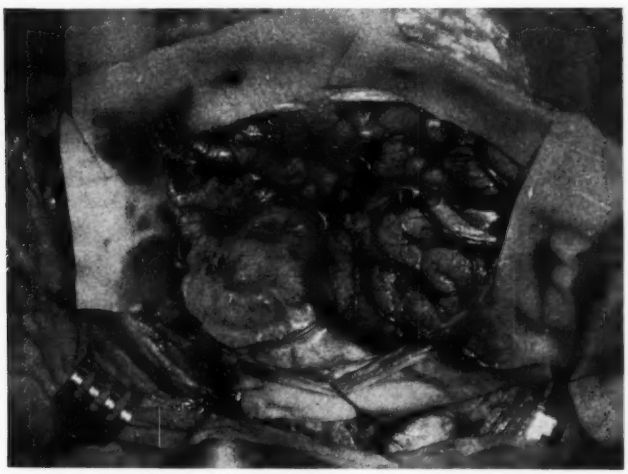

FIG. 4. *Case B.* View of operation, showing the temporal pole to be absent. The insula can be seen. There is no arachnoid covering to the underlying brain.

(Fig. 3). There was forward bulging of the middle fossa that was best seen on base views. Left carotid arteriography gave no filling of the anterior cerebral artery. The middle cerebral artery was elevated throughout its course. There was an avascular area between the brain and the skull (Fig. 3). EEG showed low voltages in the left frontoparietal leads.

Operation. A temporoparietal bone flap was elevated. The bone was very thin and bulged outwards in its centre. The dura mater was bluish and on opening it there was a large collection of clear fluid in the anterior part of the middle fossa. Inspection showed that the arachnoid had been opened with the dura mater. It was reflected for a considerable distance from the underlying frontal and temporal lobes. The arachnoid was thickened where it resumed its normal relations with the pia mater. The temporal pole of the brain was absent. The gyri were free of arachnoid and had a normal pattern, and the vessels stood out clearly. The insula was visible and the temporal operculum was absent (Fig. 4). Cerebrospinal fluid welled into the cyst from over the tentorium. The internal carotid artery, the middle cerebral artery and the oculomotor nerve were free and ran in the medial wall of the cyst. Some of the peripheral dura mater and arachnoid were taken for examination. The wound was closed and the bone flap was wired into place.

Postoperative course was satisfactory and he was well 9 months later.

Histological Examination. The membranes showed a normal dura mater and an inner membrane similar to arachnoid.

Comment. The essential feature of these 2 cases was the asymmetry of the skull without other symptoms. On inspection, the anterior part of the middle fossa was occupied by a collection of clear fluid in the subarachnoid space. The temporal pole of the brain was absent and there was no evidence of compression of the brain. Case B had the insula uncovered.

Fifteen similar cases were found in the literature (Table 1).

ANALYSIS OF MATERIAL

Analysis of the 15 cases from the literature with the 2 cases presented herein shows the common pattern.

Age and Sex. Seven cases were in the first decade, 6 in the second decade and 4 patients were over 21 years of age. There were 13 males and 4 females.

Symptoms. The left side of the head was involved in 12 of the 17 cases. In most patients the bulging of the temporal region of the skull had been evident for many years and other symptoms were trivial. Neurological signs were mostly absent or trivial. There were 4 cases (Cases 1, 7, 11 and 14) in which the symptoms were unusual. This was because of the superimposition of other factors. Cases 1 and 11 had had recent head injuries and Case 7 had malignant changes in the lining membrane. The cause of symptoms in Case 14 is uncertain. There was often a distant minor head injury and this seems to have drawn attention to the asymmetry of the skull.

Radiography. There was thinning and bulging of the temporal squama of the skull and the adjacent parts of the frontal and parietal bones were often affected. Only in Case 1 was the skull normal. In most instances the middle fossa was enlarged forwards and the lesser wing of the sphenoid was elevated.

TABLE 1

Cases of intracranial collections of fluid reported in the literature

Case No.	Author Surgeon	Age Sex Side	Symptoms and Signs	Radiology	Operative Findings
1	Cohen[3] Elsberg	20 yrs. M L	Head blow boxing 6 wks. before. Then headaches, vomiting. Papilledema, R-sided weakness.	Skull normal. Air studies by direct injection: large frontal cyst.	Trephine: 300 cc. brown fluid under dura mater. Frontoparietal flap: absent brain ant. fossa and ant. part middle fossa; II and III nerves seen. Recovery.
2	Hardman[7] (Case 3) Jefferson, Rowbotham	5 yrs. M L	11 mos. minor head injury. 9 mos. bulge L forehead.	Thinned, expanded L frontal and temporal area. Sphenoid ridge not seen on L.	Frontal trephine: 250 cc. clear fluid. Compressed frontal lobe and orbital plate seen. Brain covered with arachnoid. Recovery.
3	Bull[1] (Case 10) Olivecrona	4 yrs. M L	Barber noticed L temporal swelling.	L temporal bulge; thin bone. Air studies: L temporal horn backwards and upwards.	Temporal flap: subdural hygroma. Recovery. Well 7 yrs. later.
4	Bull[1] (Case 11) Olivecrona	7 yrs. M L	Hit by stone at 3 yrs. Swollen L temple since.	Temporal and frontal bulge; thin bone. Air studies: L temporal horn back and up, septum pellucidum slightly to R.	Temporal flap: subdural hygroma. Recovery. Well 6 yrs. later.
5	Bull[1] (Case 12)	18 yrs. M R	Lifelong headaches, temporal swelling.	Expansion middle fossa. Air studies: R temporal horn backwards.	Operation refused.
6	Petit-Dutaillis *et al.*[11] (Case 2)	12 yrs. M R	Head injury at 3 mos. Headaches, R temporal swelling, 1 yr. R VI palsy, depressed corneal. EEG unstable.	Expansion middle fossa; thin frontal, temporal, parietal areas.	Subdural hygroma in sylvian fissure. Frontal lobe spread apart. Insula and sylvian vessels seen. Recovery.
7	Pertuiset and Berdet[10] (Case 1)	32 yrs. M L	2 wks. headaches, R-sided weakness. R hemiplegia, L temporal bulge. EEG: delta waves L temporoparietal leads.	Enlarged temporal fossa. Arteriogram: sylvian vessels raised; ant. cerebral artery to R.	Xanthochromic fluid in subdural space. Cavity in temporal fossa; free edge of tentorium seen, Broca's area depressed. Membrane malignant.
8	Childe[2] (Case 2) Penfield	8 yrs. F L	Attack unconsciousness at 20 mos. Exophthalmos 3 yrs. Swelling L eye 2 yrs. L proptosis, exophthalmos 5 mm., internal strabismus.	Downward bulge middle fossa. Broad superior orbital fissure.	Temporal flap: cyst under temporal lobe from orbit to petrous ridge. Yellow fluid, 21 mg.% protein. 2nd op. 14 mos. later: similar findings.
9	Childe[2] (Case 3) Penfield	19 yrs. F R	Swelling R frontoparietal region since birth. Headaches 8 mos. R temporal bulge.	Enlarged middle fossa. Raised lesser wing sphenoid. Thin, bulging temporal, frontal, parietal bones.	Temporal flap: cyst in subarachnoid space pushing temporal lobe from sylvian fissure. Insula open.
10	Childe[2] (Case 4) Cameron	24 yrs. F L	Swelling L frontoparietal area since age 8 yrs. L side head prominent.	Enlarged middle fossa. Thin, bulging frontal, parietal areas.	Large cyst compressing frontal, temporal lobes. No lining membrane. Fluid: 25 mg.% protein.
11	Childe[2] (Case 5) Parkinson	21 yrs. M L	Minor head injury at age 3 yrs., when swelling noticed. Recent twist to head. Bulge L temple.	Middle fossa bulging forwards; bulging frontal, temporal bones. Arteriogram: middle cerebral up and medial; ant. cerebral to R and posterior.	Bipolar hygroma of ant. and middle fossae. Tentorium, II, III, V nerves, both carotids seen.
12	Childe[2] (Case 7) Walker	9 yrs. M L	L temporal bulge many yrs. Recent acute head injury. L temporofrontal bulge.	Middle fossa expanded, lesser wing sphenoid raised. Thin bulging frontal, temporal, parietal bones.	Cystic cavity between frontal and temporal lobes. Arachnoid lining. Sylvian vessels free.
13	Childe[2] (Case 8) Walker	6 yrs. F R	Headaches, R temple swelling, 3 mos.	Middle fossa slightly enlarged. Thin bulging frontal, temporal, parietal bones.	Large subdural cyst. Brain displaced along sylvian cleft. Lesser wing sphenoid seen.
14	Childe[2] (Add. Case 1) Parkinson	58 yrs. M R	Headaches, ataxia, tinnitus, olfactory hallucinations, 4 mos.	Forward enlargement R middle fossa. Raised lesser wing sphenoid. Air studies: ventricles 4 mm. to L; temporal horn backwards.	125 cc. clear fluid in cyst ant. part middle fossa. Separate membrane. Fluid: 25 mg.% protein.
15	Childe[2] (Add. Case 2) Parkinson	13 yrs. M L	Recent head injury. L temporal bulge for years.	Bulge L temporal squama. Air studies: ventricles 5 mm. to R; temporal horn posterior.	100 cc. hygroma ant. part middle fossa. Fluid: 15 mg.% protein.

In cases in which air studies of the cerebral ventricles were done (Cases 3, 4, 5, 14 and 15) there was a minimal shift of the ventricles to the side opposite the bulge. The temporal horn was truncated or dorsally displaced. In none was the cyst filled with air. Carotid arteriography (Cases A, B, 7 and 11) showed that the middle cerebral vessels were displaced upwards and medially while the ipsilateral anterior cerebral artery was shifted to the opposite side.

Electroencephalography. This was done in Cases A, B, 6 and 7 and in 3 of the 4 there were focal features.

Fluid Contents. In 5 cases (Cases 2, 8, 10, 14 and 15) the cyst fluid was analysed for protein content and it was similar to the cerebrospinal fluid findings; in no instance was the protein content raised.

Operative Appearances. The common feature was that the anterior part of the middle fossa was occupied by a large collection of clear fluid—up to 125 cc.—so that a gap appeared between the frontal and temporal lobes. The sylvian fissure was opened up. The insula was uncovered in 3 cases (Cases B, 6 and 9) and probably so in Case 7. Cases 10 and 11 had absence of the polar portions of the frontal lobe as well, and Case 1 had a frontal polar cyst but the state of the temporal lobe was not known. The free edge of the tentorium was often seen and a variety of cranial nerves from the olfactory nerve to the gasserian ganglion were seen without retraction of the brain. The internal carotid artery and its major branches were exposed.

From many of the operative accounts it is difficult to decide what were the exact relations of the fluid collections to the containing membranes. The trend of description is to suggest that the fluid was in the subdural space. The fluid was subarachnoid in Cases A and B. There was a separate lining to the cyst in Cases 6, 10, 13 and 14 and there was arachnoid between the cyst wall and the brain.

Results. Case 5 was not operated upon. The rest of the patients recovered from their operations and were well for periods up to 7 years. No mention was made of any alteration in bulging of the skull.

DISCUSSION

Penfield provided 2 cases for Childe's series[2] and in the subsequent discussion he said,[9] "In the past I, like some others, have assumed that the fluid was subdural but I am beginning to wonder whether in these cases that he has presented the collection may not be subarachnoid instead." No further evidence was offered. This point is exemplified by the literature. Surgeons have usually believed that no large collection of fluid could occur in the subarachnoid space because of its trabeculation. Any large fluid mass would be deemed to be in the subdural space which has a great potential capacity. It is not easy to determine the exact relations of the membranes when a limited exploration of the intracranial contents is made. We were sure of the subarachnoid location of the fluid in our cases and there were no additional membranes.

These collections of fluid have been called subdural hygromas by most

authors. This has carried with it the implication that they are related to the subdural hygromas that follow a closed head injury. A minor head injury was believed to have initiated the condition in several cases. However in others the bulging of the skull had been noticed at an early age and an injury served to accentuate the asymmetry of the skull. It is probable that this condition is determined prenatally and any subsequent injury is a matter of coincidence.

Some operative accounts suggested that the brain was displaced by the fluid to such an extent that the tentorium and its related structures were visible. Yet clinical disturbances were trivial. It is not likely that such displacements would be compatible with life. In our cases the brain was not displaced or compressed and the gyral pattern was normal. Our impression was that the temporal pole was absent.

Cunningham[5] has described the development of the brain in man. The temporal lobe is formed from a caudal bulge of the hemisphere that grows in a downwards and forwards direction. At first there is a sylvian depression between the temporal lobe and the rest of the hemisphere and this deepens to become the sylvian fissure. At about 6 months the sulci and insula appear. In the next 3 months the insula is covered by the opercula growing over from the frontal, frontoparietal and temporal regions. The insula is opercularised by birth.

If it is postulated that this condition is caused by failure of the temporal lobe to come forwards in its development then the temporal operculum will be absent and the lower part of the insula uncovered. This was the state of affairs in Cases B, 6 and 9 and probably in Case 7. Air studies showed the lack of forward position of the temporal horn of the lateral ventricle. The ease with which the structures on the medial side of the temporal lobe can be seen, can be best accounted for by a temporal lobe agenesis. A grosser degree of the deformity would be when the frontal pole fails to develop as well.

It is suggested that this condition of a fluid collection in the region usually occupied by the temporal pole of the brain and associated with enlargement of the boundaries of the middle cranial fossa is caused primarily by agenesis of the temporal lobe. It may co-exist with agenesis of the frontal lobe. The fluid is cerebrospinal fluid. It cannot be denied that this agenesis could be determined by a pre-existing cyst at this site. Our evidence points to a subarachnoid location for the fluid but in some cases there seems to have been a thin extra membrane. It is not considered that this condition is related to the arachnoidal cysts that occur elsewhere in the nervous system. These act as progressive space-occupying processes with corresponding clinical effects. With the material under discussion there is some exceedingly slow expansion of the skull that stabilises at adult life but there are no other symptoms in the uncomplicated case.

It is less easy to account for the expansion of the skull. It is well known that most types of agenesis of the brain are associated with a corresponding lack of development of the skull on that side. Intracranial tumours and

hematomas tend to bulge the skull over them in patients under the age of 15 years,[8] but in most of these cases there is the added factor of raised intracranial pressure. Pascal's law states that in a fluid, the pressure applied at any point is transmitted equally in all directions. Yet with these pathological conditions there is some local escape from this law. Connolly[4] has suggested that the digital impressions of the skull in adolescence and in hydrocephalic states result from a closer contact of the brain surface with the inner table of the skull than previously. Close tissue proximity certainly occurs with most hematomas and tumours, and produces, perhaps, what are no more than gigantic digital impressions. Somehow or other the fluid collections in temporal lobe agenesis act similarly although the dynamic factors are far from clear.

DIAGNOSIS AND TREATMENT

Other causes of localised bulging of the skull in childhood and adolescence are skull tumours, chronic subdural hematomas and cerebral neoplasms. The skull tumours can be readily distinguished by their radiological appearances. The chronic subdural hematomas have neurological manifestations that they share with cerebral tumours. The usual radiological appearances of the subdural hematomas[1,6] are elevation of the lesser wing of the sphenoid, bulging out of the greater wing of the sphenoid and bulging forwards of the middle fossa. Similar changes may occur with a temporal lobe glioma and are present in temporal lobe agenesis. Air studies and electroencephalography may be inconclusive. Carotid arteriography will be helpful. There will be an avascular area between the brain and the skull in cases of hematomas and in many cases of temporal lobe agenesis, but the upwards course of the middle cerebral artery will probably be absent with hematomas. With a temporal lobe neoplasm the middle cerebral artery would be elevated but there would be no avascular space between the brain and the skull. The upwards course of the middle cerebral artery is most noticeable in its first part with the agenesis and greater than with a tumour.

It is suggested that the patient in the first two decades of life who has bulging of the temporal region of the skull that is not associated with any other obvious clinical features, have the skull radiographed and nothing further done. A short period of observation will soon confirm the simple nature of the lesion. There is no evidence that surgical exploration influences the course of the condition. When there are additional clinical signs then full investigation and perhaps a craniotomy are needed.

SUMMARY

Two cases have been presented in which there was bulging of the skull without other physical signs. At operation the temporal pole of the brain was absent and its place was occupied by a large amount of cerebrospinal fluid.

Fifteen similar instances have been found in the literature under the designation of subdural hygromas.

It is suggested that these are cases of temporal lobe agenesis. Agenesis of the frontal lobe may exist as well.

The diagnosis is considered but no treatment is advised.

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CRANIOPHARYNGIOMA (PITUITARY ADAMANTINOMA) IN PATIENTS MORE THAN 60 YEARS OF AGE

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CRANIOPHARYNGIOMAS are thought to arise from embryonic cell rests and, therefore, to be congenital in origin.[5,11,14] The majority of these tumors occur in childhood or early adult life,[4,12–14] and it is unusual for one to occur in a person more than 60 years of age.[13] In 10 different series assembled from the literature, we were able to find reports of 454 craniopharyngiomas, only 5 of which were in persons more than 60 years of age (Table 1).

We wish to report on two patients with craniopharyngioma who were more than 60 years of age and who were operated on at the Mayo Clinic in the last 2 years. One was 70 years of age at the time of operation, and is, to the best of our knowledge, the oldest person ever reported as having this type of tumor.

REPORT OF CASES

Case 1. A 70-year-old white woman, referred to the Mayo Clinic because of failing vision, said that she had been in good health until 3 months prior to admission on January 8, 1954, when she first noticed difficulty in seeing to thread needles and in doing close work such as required in sewing. She thought that she could see better when looking straight ahead than when looking to the sides. This difficulty in vision steadily increased until at the time of admission she could distinguish only large objects with the right eye and had difficulty in seeing objects to the side with the left one. In recent months she had experienced occasional headaches localized to the vertex. These came on early in the morning and lasted a short time; they had not incapacitated her.

The past history was uninformative. She had been in excellent health all her life. She had been pregnant nine times and had seven living children. There were no symptoms suggestive of pituitary insufficiency or diabetes insipidus. She had weighed 190 lbs. (86.2 kg.). Twenty years ago and at the time of admission she weighed 200 lbs. (90.7 kg.).

Physical examination disclosed an obese white woman. The blood pressure measured 168 mm. of mercury systolic and 88 mm. diastolic. The pulse rate was 92 beats per min. and the temperature was normal. The distribution of hair was

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TABLE 1

Cases of craniopharyngioma reported in the literature: total patients and patients more than 60 years of age

Author	Patients	
	Total	More than 60 Years of Age
Duffy[5] (1920)	55	—
Critchley and Ironside[3] (1926)	7	1
Beckmann and Kubie[1] (1929)	16	—
Frazier and Alpers[6] (1931)	14	—
Cushing[4] (1932)	92	2
Carpenter *et al.*[2] (1937)	12	—
Globus and Gang[7] (1945)	14	—
Grant[9] (1948)	48	?
Gordy *et al.*[8] (1949)	51	1
Müller and Wohlfart[14] (1950) (Olivecrona series)	45	—
Love and Marshall[12] (1950)	100	1
Total	454	5

normal, and there was no abnormal pigmentation of the skin. General examination gave negative results except for the visual-field changes shown in Fig. 1a.

Roentgenograms of the thorax showed pleural thickening in the left base. A

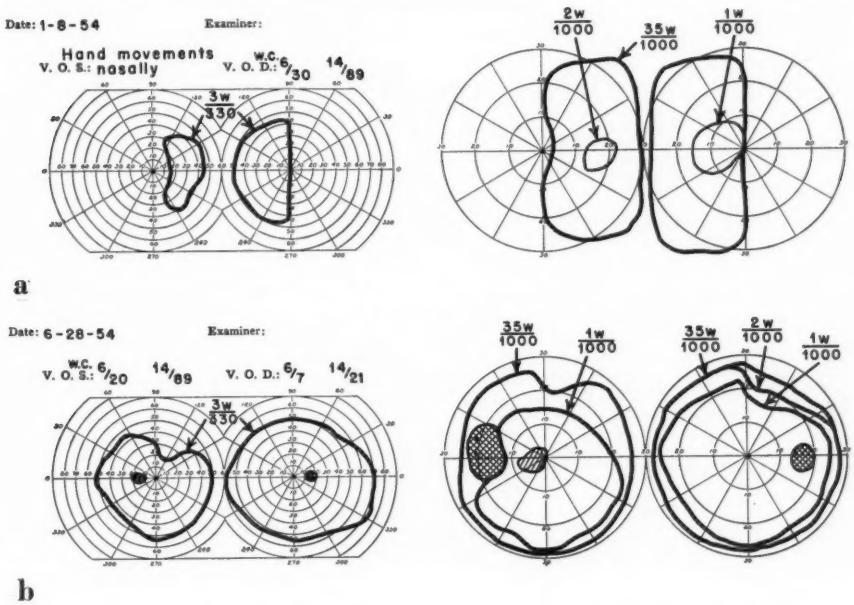

FIG. 1. Visual fields in Case 1 before operation (a) and 6½ months postoperatively (b).

roentgenogram of the skull disclosed decalcification of the posterior clinoid processes. The pineal calcification was in normal position.

Urinalysis disclosed a specific gravity of 1.017, albuminuria, grade 1 (on a basis of 1 to 4), and pyuria, grade 3. The hemoglobin measured 13.2 gm. per 100 cc. of blood, and the leukocyte count was 9,600 per c.mm. The urinary 17-ketosteroids measured 3.1 mg. and the urinary corticosteroids 0.55 mg. per 24 hours. The basal metabolic rate was +7 per cent.

The patient was given 200 mg. of cortisone intramuscularly 48, 24 and 2 hours before operation. On January 18, 1954, a frontal craniotomy was done. The chiasm was approached over the right frontal lobe. A solid, microcystic adamantinoma was found presenting between the optic nerves. The tumor was extensive, but could be delivered from under each optic nerve and from the intact pituitary body without difficulty. The apex of the tumor was adherent to the hypothalamus; after debate it was decided to leave this portion of the tumor for fear of hypothalamic damage if attempts were made to remove it. The wound was closed without drainage.

The postoperative course was uneventful. Since the patient had shown no evidence of pituitary insufficiency, either before or after operation, the daily dose of cortisone was gradually decreased, being discontinued on the 4th postoperative day. The wound healed without incident and the patient was permitted to go home on the 13th postoperative day.

Twenty-four days later (37 days after operation) she was readmitted to the hospital because of anorexia, nausea, vomiting and weakness. She had felt well for the first 2 weeks after returning home, but then began to notice anorexia and easy fatigue. One week prior to readmission she had begun to have periods of nausea and vomiting, beginning about an hour after eating. She also had had hiccoughs for short intervals two or three times per day for the past week. She denied salt-craving, excessive thirst or urinary frequency, and had no episodes of fainting or collapse, but had lost 16 lbs. (7.3 kg.) since returning home.

Examination revealed a blood pressure of 140 mm. of mercury systolic and 70 mm. diastolic. The pulse rate was 90 beats per min. The skin was dry and there was slight pigmentation in the axillary folds and inguinal regions. The carbon dioxide-combining power measured 26 mEq. per liter of plasma; serum chlorides, 100.3 mEq. per liter; urinary 17-ketosteroids, 0.5 mg. per 24 hours; urinary corticosteroids, 0.20 mg. per 24 hours; hemoglobin, 12.7 gm. per 100 cc. of blood; erythrocytes, 4,050,000 per c. mm.; and basal metabolic rate, −4 per cent.

The patient was started on treatment with fluids by vein and cortisone in doses of 10 mg. every 8 hours. She quickly regained her appetite and after the first 2 days had no further nausea or vomiting. She had no signs or symptoms of myxedema at any time and, with marked improvement, was dismissed after 12 days to return home, taking cortisone in maintenance doses of 5 mg. three times a day.

Re-examination 3 months later, on June 28, 1954, revealed excellent progress. She had an excellent appetite, was able to be up and about all day, and was able to work about the house. The basal metabolic rate was −13 per cent on this occasion. The urinary 17-ketosteroids measured 0.4 mg., and the urinary corticosteroids 0.54 mg., per 24 hours. There were no stigmata of myxedema. Examination of the perimetric visual fields (Fig. 1b) showed gratifying return of vision.

Case 2. A 63-year-old white woman first visited the Mayo Clinic early in September, 1952, because of failing vision in the left eye. This had first appeared about

9 months previously. She had had several brief headaches during the past 9 months, but these had never been severe enough to require aspirin for relief. One month previously, she experienced two short episodes of diplopia. She consulted her local physician, who referred her here for further study.

She had been in excellent health all her life. She had four living children and had had three miscarriages. She had undergone surgical repair for procidentia 14 years previously.

Physical examination gave normal results in all respects, with the exception of a small cystocele. She was 5 ft. and 5 in. (165.1 cm.) tall, and weighed 150 lbs. (68 kg.). The blood pressure measured 132 mm. of mercury systolic and 78 mm. diastolic. The pulse rate was 64 beats per min., and the temperature was 98.2°F.

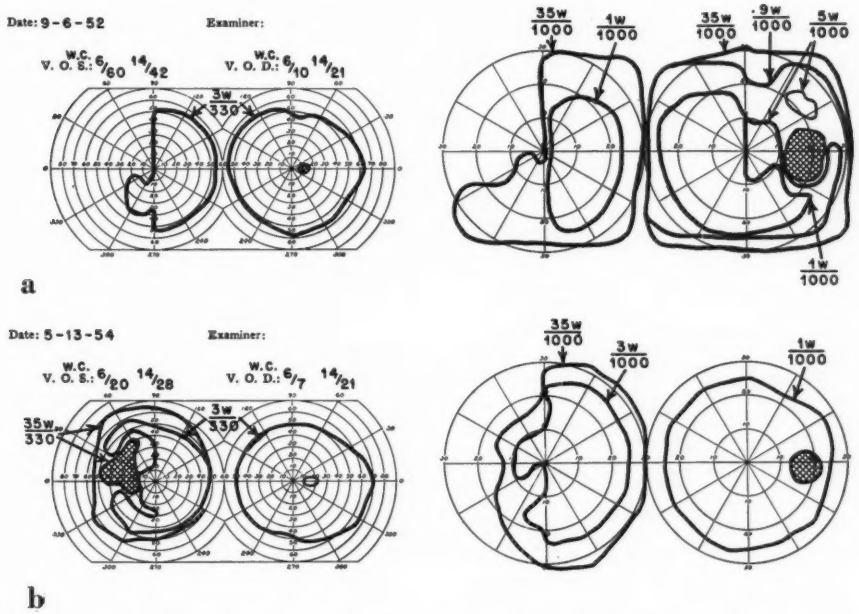

FIG. 2. Visual fields in Case 2 before operation (a) and 20 months after operation (b).

The visual fields are shown in Fig. 2a. There were no clinical signs of pituitary insufficiency.

The hemoglobin measured 12.6 gm. per 100 cc., and the white blood count was 5,200. The urine had a specific gravity of 1.017, but was otherwise entirely normal. The urinary 17-ketosteroids measured 1.9 mg. per 24 hours. The basal metabolic rate was −11 per cent. A roentgenogram of the thorax did not disclose any abnormality. A roentgenogram of the skull revealed thinning and decalcification of the dorsum and floor of the sella. The patient was given 200 mg. of cortisone intramuscularly 48, 24 and 2 hours preceding operation.

On September 24, 1952, a left frontal craniotomy was done. The chiasm was exposed through an intradural approach and a cyst was encountered lying under the

optic nerves and the optic chiasm. Aspiration disclosed that the cyst was filled with clear cerebrospinal fluid. This aspiration seemed to decompress the chiasm. Further exploration revealed a solid tumor underlying the hypothalamus. This was resected and proved to be a solid pituitary adamantinoma. A small portion of the capsule of the tumor, which was adherent to the carotid arteries, was not removed as it was thought that the risk was too great in view of the age of the patient. The wound was closed without drainage.

The postoperative course was uneventful, and since there were no signs of pituitary insufficiency either preoperatively or postoperatively, the dosage of cortisone was decreased gradually and administration was discontinued on the 7th postoperative day. Mild diabetes insipidus developed, but this was easily controlled with nasal insufflation of posterior pituitary powder three or four times a day. The patient was dismissed to return home on the 12th postoperative day, without medication other than the posterior pituitary powder.

Sixty-five days after operation, the patient was readmitted, complaining of listlessness, apathy, anorexia, nausea and vomiting. She said that she had felt listless since arriving home from the hospital, and that her skin and mouth had been dry all the time. She was able to be up and about most of the day. One week before readmission, her local physician found her blood count low and gave her blood by transfusion. This was followed by nausea and vomiting once or twice per day, as well as by anorexia. She had also had occasional episodes of hiccoughing. She had not fainted or collapsed at any time.

Physical examination revealed a blood pressure of 117 mm. of mercury systolic and 70 mm. diastolic. The pulse rate was 80 beats per min. The temperature was normal and the skin was dry. The distribution of hair was normal, and there was no abnormal pigmentation. The value for hemoglobin was 13.8 gm. The erythrocyte count was 5,040,000, and the leukocyte count 5,100. Urinalysis gave normal results. The value for plasma chlorides was 97.0 mEq. per liter; carbon dioxide-combining power, 17.9 mEq. per liter; serum sodium, 128 mEq. per liter; serum potassium, 3.7 mEq. per liter; urinary 17-ketosteroids, 0.2 mg. per 24 hours; and urinary corticosteroids, 0.24 mg. per 24 hours.

Cortisone, 12.5 mg. by mouth every 8 hours, was prescribed and, in addition, the intravenous administration of fluids was started. The condition of the patient improved rapidly, and after the 2nd day she had no further nausea or vomiting. Her appetite was good. The amount of cortisone was gradually decreased to a maintenance dose of 5 mg. given three times a day. The diabetes insipidus continued to be mild and was readily controlled with three to four nasal insufflations of posterior pituitary extract per day. The basal metabolic rate was −5 per cent, and there was no clinical evidence of myxedema. Accordingly, desiccated thyroid was not prescribed.

When the patient was examined, 20 months after operation, she was in excellent health. She was able to do all her housework without undue fatigue, and lived a normal life. She continued to take cortisone in doses of 5 mg. three times a day, and nasal insufflations of posterior pituitary powder three times a day. There were no signs or symptoms of thyroid deficiency. Examination of the perimetric or visual fields on May 13, 1954, showed gratifying return of vision (Fig. 2b).

COMMENT

It is always difficult to decide whether complete removal of these tumors should be attempted. Complete removal is the only certain way of pre-

venting recurrence, but technically it is exceedingly difficult and at times impossible without causing death of the patient. Aspiration of the cyst and subtotal removal of the capsule is frequently followed by early recurrence of symptoms, although some surprisingly long survivals have been reported. In both of the present cases it seemed impossible to remove the last vestige of the tumor without inviting disaster by injuring the carotid arteries in one instance, or the hypothalamus in the other.

To date, 10 months and 24 months respectively after operation, there has been no evidence of recurrence, and the striking return of vision has been very gratifying in our cases.

The evaluation of the endocrine status in patients with this type of tumor may be difficult. A considerable proportion of such patients appear clinically to be quite well, and without evidence of adrenal, pituitary, thyroid or gonadal disorder, yet it is possible that many of them may have some minor degree of impaired glandular function.[10] A minority of the patients present, when first examined, clear-cut pictures of panhypopituitarism with a characteristic pale, waxy color, scantiness of the axillary and pubic hair, absence of hair or the presence of very fine body hair, dryness and thinness of the skin, thinness and weakness of the muscles, and asthenia, with or without signs of pituitary myxedema. In all cases in which pituitary tumor is found, whether or not such symptoms are observed, it is well to evaluate thyroid, adrenal and, if possible, gonadal function with such laboratory aids as are available.

In recent years it has been our practice, however, to prepare with adrenal substitution therapy all patients who are to undergo an operation for pituitary tumor, regardless of whether physical examination or laboratory study shows any evidence of pituitary or adrenal insufficiency. This program is difficult to evaluate objectively since many patients have tolerated pituitary operations uneventfully without such prophylactic adrenal replacement therapy. It is nevertheless our impression that this regimen has resulted in substantial improvement in the postoperative course and subsequent results in this group of patients.* One reason, of course, why such treatment might be helpful, even in the absence of presurgical pituitary or adrenal insufficiency, is related to the fact that manipulation of the pituitary at operation may impair its ability to elaborate corticotropin (ACTH).

While theoretically either cortisone or ACTH might be satisfactory for use as a prophylactic agent for such patients before operation, we have generally preferred to use the former. It is our practice to prepare all patients with intrasellar or suprasellar tumors, regardless of the presence or absence of pituitary insufficiency, with cortisone in doses of 200 mg. always given intramuscularly 48, 24, and 2 to 4 hours before operation. The object of this treatment is to provide the patient with several large depots of adrenal steroid on which he or she may draw during and after operation, when stress is likely to be a serious factor. The dose indicated was chosen because

* An analysis of our results with this program is being made by Troen and Rynearson and will be published elsewhere.

it was considered very likely to be maximal and probably more than the largest dose that was really needed.

Neither of the two patients presented in this report exhibited preoperative pituitary or adrenal insufficiency. The postoperative treatment in such cases is gauged by the patient's response. The dosage of cortisone is usually reduced rapidly unless a demonstrated need is observed, and administration is often discontinued within the first week. The fact that this sort of program was followed in our two cases may have contributed in some measure at least to the uneventful course that was observed immediately postoperatively.

The episodes of weakness and collapse that appeared 37 and 65 days respectively after operation are presumed to have been a result of adrenal insufficiency. This presumption is based on the history and presenting symptoms in each case and on the clear-cut response to treatment with cortisone, although the clinical and chemical findings in both episodes were essentially negative. Such occurrences of adrenal insufficiency have been unusual in our experience and are not readily explained. More commonly, such episodes occur abruptly soon after operation and in response to some intermittent stress, but such precipitating factors were not evident in the present instances. These experiences may provide support for the view that some continuous adrenal replacement therapy may be desirable after any operation on the pituitary body, even when objective signs of overt pituitary insufficiency are lacking.

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THE COLLATERAL CIRCULATION FOLLOWING MIDDLE CEREBRAL BRANCH OCCLUSION

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ARTERIOGRAPHIC demonstration of occlusion of the main stem of the middle cerebral artery has been reported in a number of instances,[1,6–10] and the peripheral collateral circulation following such occlusion has been illustrated on several occasions.[6,8–10] The criteria for demonstration of these features are (1) failure of the middle cerebral vessel to fill beyond a point of obstruction, and (2) delayed retrograde filling of the cortical branches of the middle cerebral artery from branches of anterior and posterior cerebral arteries. The arteriographic features of middle cerebral branch occlusion have not received so much attention. The following is presented because of the interesting pattern of collateral circulation observed and also because the events illustrate some features of the natural history of cerebral vascular disease now recognized to be important.

CASE REPORTS

Case 1. The patient was a 36-year-old man who had been healthy apart from attacks of malaria contracted in 1943 when overseas and continuing until 1947. In 1948, he began to experience sudden episodes of dizziness and blurring of vision, with numbness around his mouth on both sides and inability to speak. By 1950, the attacks also included weakness and numbness of the right arm and leg, lasting from several minutes to half an hour, and followed by general malaise for several weeks. From the onset, the average frequency of attacks was one per month, although in the 5 months prior to admission he had been quite free.

On June 25, 1954, the patient went to bed feeling well. His wife awakened at 7:30 A.M. June 26, and saw the patient rubbing his right arm with the left hand and moving his tongue about in his mouth. He was unable to speak or to use the right arm and leg. He was admitted to the Denver Veterans Administration Hospital that morning.

Examination. The ocular fundi and eye movements were normal. There was inattention to objects in the right field of vision, but in answer to direct questioning he indicated that he did see objects in that field. There was profound right hemiparesis, together with depressed tendon reflexes and an indefinite plantar response on that side. The right superficial abdominal reflexes could not be elicited. He was unable to speak, and though unable to respond to verbal commands did appear to understand some of what was said to him. Blood pressure was 125/80. Plain radiographs of the skull revealed no abnormality. It was thought that there might be a

left internal carotid artery occlusion, but the possibility of a bleeding vascular anomaly in the left hemisphere was also considered.

Angiography. On the afternoon of admission, left carotid angiography was carried out. For the anteroposterior film, 8 cc. of 35 per cent diodrast were injected, and 4 exposures were made at 1-sec. intervals. Subsequently, two further injections were made for stereoscopic lateral series, the time intervals being the same.

On the first film of the anteroposterior series (Fig. 1), made just before the completion of the dye injection, there was seen to be obstruction of the superior of the two visible branches of the middle cerebral artery, 2 cm. from the carotid bifurcation. In addition, there was noted a paucity of vessels in the peripheral distribution

FIG. 1. *Case 1.* There are two visible branches of the middle cerebral artery, one of which (A) is obstructed. Note the paucity of vessels in the peripheral area of supply of the middle cerebral artery (B).

of the ascending branches. On the first film of the lateral series (Fig. 2) there was good filling of the posterior temporal branch, but no filling in the territory of distribution of the ascending frontoparietal or parietal branches. On the second film, made 1 sec. later (Fig. 3), reflux filling of these branches from internal frontal, paracentral, and precuneal branches of the anterior cerebral artery was shown. This filling persisted into the next frame (1 sec.).

Course and Treatment. Immediately after arteriography, the patient's paralysis became worse, but several hours later it had returned to the pre-arteriography level. He continued to improve in strength, and his speech returned. He left the hospital, and has not responded to requests to return for examination.

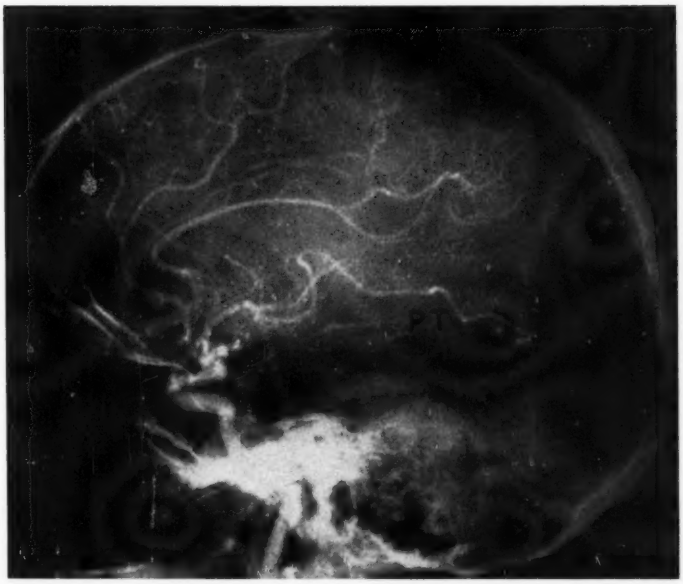

FIG. 2. *Case 1.* Filling of the posterior temporal branch of the middle cerebral artery (PT) but absence of filling in the area of supply of the ascending branches.

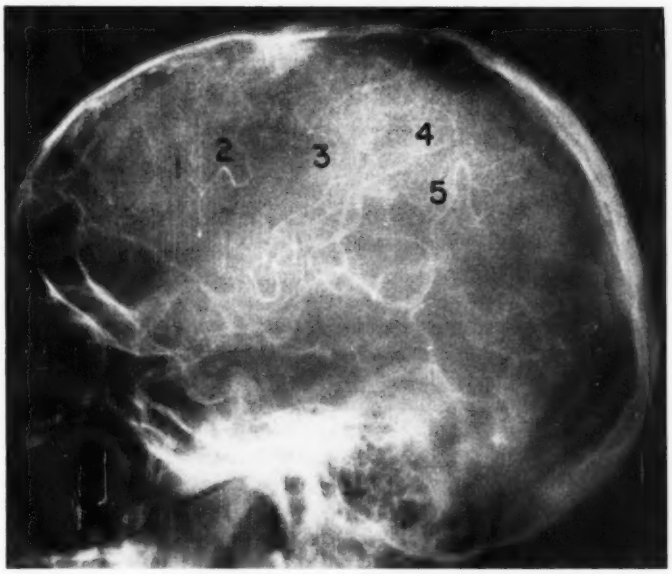

FIG. 3. *Case 1.* Late reflux filling of the ascending branches (1, 2, 3, 4, and 5) from branches of the anterior cerebral artery.

Summary. A 36-year-old man had experienced intermittent attacks of aphasia and right hemiparesis with sensory disturbances for 6 years. He awakened one morning with severe aphasia, right hemiplegia, and inattention in the right visual field. Angiography showed obstruction of a primary branch of the left middle cerebral artery, which was the common origin of the ascending branches. The posterior temporal branch was spared, but the collateral circulation into the ascending frontoparietal and parietal arteries occurred, not from the posterior temporal artery, but instead from branches of the anterior cerebral artery.

FIG. 4. *Case 2.* The most proximal part of the middle cerebral artery is filled, but just beyond the origin of the posterior temporal artery (PT) there is obstruction (arrow).

Case 2. A 47-year-old man, who had had a traumatic amputation of the left leg at the mid-thigh in 1928, was admitted to Denver General Hospital on Sept. 16, 1954, complaining of paralysis of his left arm and the stump of his left leg. Except for syphilis contracted many years before, there were no previous illnesses of note. His present complaint dated from 2 days prior to admission, when he awakened after about two hours' sleep, feeling light-headed and noticed that he was unable to move his left arm or the stump of his leg. His tongue felt thick and awkward, resulting in some difficulty in speech, but his words and sentences were accurately formed. During the subsequent 2 days at home, the difficulty in speech almost disappeared, and there was considerable return of function in the stump, but his left arm remained useless.

Examination. He was an alert and cooperative patient in no distress, with a normal temperature, pulse, and respiratory rate, a blood pressure of 145/85, and no evidence of any cardiac abnormality. The positive findings were confined to the nervous system. The fundi and visual fields were normal, and the cranial nerves were intact except for a left facial weakness of central type, with protrusion of the tongue to the left. There was a flaccid paralysis of the left arm, with diminished reflexes, and weakness of flexion of the stump of the left leg. All cutaneous sensation was intact, but there was impairment of position sense and two-point discrimination in the fingers of the left hand.

In view of the previous history of syphilis and a positive VDRL in the blood, the spinal fluid was examined. It contained 1 lymphocyte, 30 mg. of protein, and the VDRL was negative.

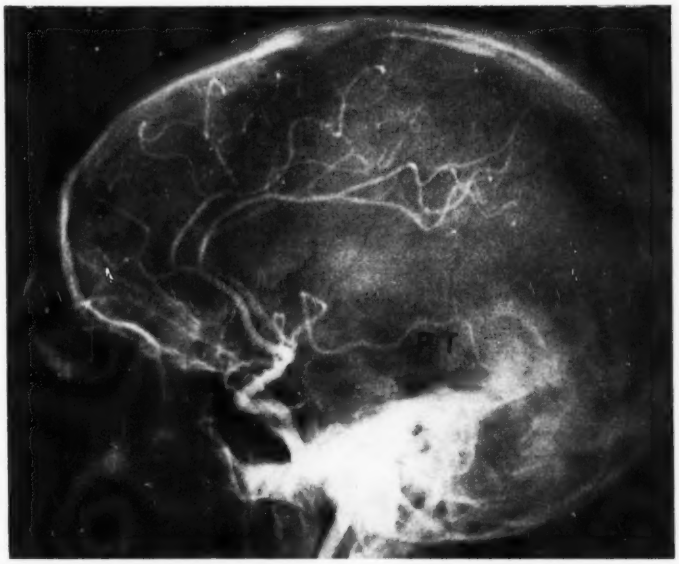

FIG. 5. *Case 2.* Note the filling of the posterior temporal branch of the middle cerebral vessel (PT) but the lack of filling in the remainder of the territory of distribution of the middle cerebral artery.

Course. By Oct. 7, 1954, considerable improvement had taken place. There was still a moderate left facial paresis, and the left arm showed a selective weakness of the extensors of the wrist, fingers, and elbow and abductors of the shoulder. Fine movements of the fingers were still very impaired and the reflexes in that limb were hyperactive. A diagnosis of thrombosis of the right middle cerebral artery was made. In view of the age of the patient, it was considered justifiable to verify this by arteriography. The patient was transferred to Colorado General Hospital on Oct. 8, 1954.

Angiography. Right percutaneous internal carotid angiography was performed on Oct. 8, 1954. Approximately 5 cc. of 35 per cent diodrast were injected, and a single anteroposterior film was made at the conclusion of the injection. Subsequently,

the procedure was repeated and a lateral film obtained. A third injection was made, and 1 sec. after the end of the injection a lateral exposure was taken. On the anteroposterior film (Fig. 4) the internal carotid and anterior cerebral arteries filled normally; the middle cerebral artery was obstructed 1 cm. distal to the carotid bifurcation and just beyond the origin of the posterior temporal branch. On the first lateral film (Fig. 5) the posterior temporal branch filled, but there was no filling of ascending frontoparietal, parietal or angular branches. The second lateral film (Fig. 6), made after a delay of 1 sec., showed retrograde filling from anterior cerebral branches. It was considered that branches 1, 2, and 3 in Fig. 6 represented frontoparietal; 4, parietal; and 5 and 6, angular branches.

Course. No untoward symptoms developed until an hour after the conclusion of

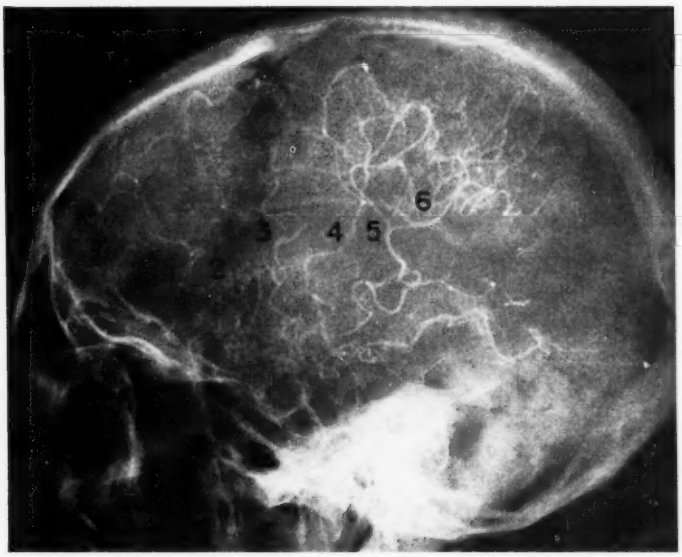

FIG. 6. *Case 2.* Delayed retrograde filling of the ascending branches of middle cerebral artery (1, 2, 3, 4, 5, and 6) from anterior cerebral branches.

the injection, when complete paralysis of the left arm was noted. During the next 4 days, steady improvement occurred. On October 12, the thrombus was excised. The demonstration of re-establishment of the circulation through its usual channels and subsequent progress will be described in a further communication.

Summary. A 47-year-old man, with no evidence of hypertension or cardiac disease, developed, without loss of consciousness, a left hemiplegia which was shown angiographically to be caused by occlusion of the common origin of the ascending branches of the right middle cerebral artery. The posterior temporal branch was spared, and the collateral circulation was established from branches of the anterior cerebral artery rather than from the open branch (posterior temporal) of the middle cerebral artery.

DISCUSSION

It is known that when a major cerebral artery (anterior, middle, or posterior cerebral) is occluded, the collateral circulation to the cortical distribution of the occluded vessel occurs through branches of the other major cerebral arteries.[2,3,6,9–11] It is interesting that when a branch of the middle cerebral artery is obstructed, the vessel distal to the occlusion fills from another major vessel rather than from the open branches of the middle cerebral artery. But this observation is not new.

Beevor,[2] in 1909, made the following experiment, utilizing a human brain removed at autopsy:

"The third branch of the middle cerebral artery, viz., the ascending parietal branch, was ligatured close to the Sylvian fissure, and the anterior, middle and posterior cerebrals were then simultaneously injected after cutting out the posterior communicating artery. It was found on subsequently cutting the hardened brain horizontally that the convolutions supplied by this ascending parietal branch were injected by the coloured gelatine from the anterior cerebral artery. This would show that either there is no communication between the contiguous branches of the middle cerebral artery, or that the communication is so slight that it is easier for the blood to reach the area corresponding to the ligatured artery from the branches of the anterior cerebral than from the contiguous branch of the same artery, or, in other words, that the communication between the middle cerebral artery and the anterior cerebral artery, where their areas touch, is much more free than that between the contiguous branches themselves of the middle cerebral artery."

Clearly Beevor's conclusion is as valid in life as in the circumstances under which his observations were made. The corollary of this is that the effect of occlusion of a cortical branch of the middle cerebral is, with regard to the cortex to which that branch is assigned, equivalent to obstruction of the main trunk of the middle cerebral artery.

Finally, the present cases illustrate important features of the natural history of "stroke." There can be little doubt that the vascular occlusion that led to the first patient's hospitalization occurred at least 6 years before, and that episodes of relative vascular insufficiency accounted for his symptoms throughout the years. This sort of vascular inefficiency has been emphasized by Denny-Brown[5] and Corday *et al.*[4] The severe disturbance which led to study of this case cannot be accurately accounted for from the data available. The recovery which was made in both cases illustrates the remarkable potential for improvement when the middle cerebral artery is occluded distal to the ganglionic branches. As has been pointed out earlier,[9,11] full credit for this must be given to the functioning of the meningeal arterial anastomoses. That dependence upon these is precarious, however, is illustrated in the clinical history of the first case.

SUMMARY

Two cases of occlusion of a primary branch of the middle cerebral artery have been observed. The occluded branch in each case was the common origin of the ascending branches.

Angiography demonstrated the lesion in both patients, and showed that collateral circulation beyond the obstruction came from anterior cerebral branches rather than from the intact part of the middle cerebral arterial tree.

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VERTEBRAL ANGIOGRAPHY BY RETROGRADE INJECTION OF THE BRACHIAL ARTERY

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VERTEBRAL angiography has become of sufficient value in the diagnosis of lesions of the posterior fossa to warrant exploring new methods to accomplish the procedure. The technique of filling the vertebral system has been described by many authors (Moniz,[3] Shimidzu,[6] Sjöqvist,[7] Takahashi,[9] Sugar *et al.*,[8] Lindgren,[2] Radner,[4] Sergent,[5] and Hauge[1]), and there are many excellent descriptions of the vascular anatomy and pathology available. At present there are three methods in general use: vertebral arterial catheterization (Radner[4]); percutaneous puncture of the artery in the neck (Sugar[8]); and percutaneous puncture of the artery at the base of the cranium (Sergent[5]). These techniques are designed primarily for use in adults and are difficult to perform in children. The catheterization technique, while described by Radner[4] in a child aged 2 years, requires considerable experience and equipment that ordinarily is not available in a radiological department. Hence there remains a need for a more adaptable method of obtaining filling of the vertebral-basilar system in infants and children.

The purpose of this report is to present our experience in a series of 16 cases in which an attempt was made to fill the intracranial vascular tree by retrograde brachial artery injection. Since 1952 this procedure has been carried out upon 9 adults and 7 children, ranging in age from 7 months to 65 years. Successful filling of the posterior fossa circulation was obtained in 5 of the 9 adults and in all of the children. In the 4 adult cases classified as unsuccessful there was some degree of contrast medium in the vessels but it was considered insufficient for diagnostic purposes.

TECHNIQUE AND ANATOMY

General anesthesia with intravenous pentothal-curare solution and endotracheal nitrous-oxide and oxygen was used on all of the children and 4 of the 9 adults. In the remaining 5 adults the anesthesia was limited to local infiltration with 1 per cent procaine.

An incision 3–4 cm. long was made over the bicipital groove at the junction of the middle and distal thirds of the arm. The segment of the brachial artery exposed at this level is distal to the origin of the profunda brachii and the superior ulnar collateral arteries. The brachial fascia was incised and the neurovascular bundle identified. The median and ulnar nerves were retracted to expose the deeper lying brachial artery. A 2–3 cm. segment of the artery was mobilized and isolated by

placing behind it a small rubber dam drain. A needle with stylet was selected according to the size of the artery. An 18-gauge spinal needle was used in the child 7 months old, while a 15-gauge needle was easily introduced in the older patients. The needle was inserted in a proximal direction for 1–2 cm. The stylet was left in place throughout the procedure except during the actual injection. A small bulldog clamp was placed on the artery distal to the entrance of the needle to prevent peripheral flow of the contrast medium. The clamp was removed between injections. The amount of the contrast medium (35 per cent Diodrast) used varied according to the size and age of the patient. A volume of 8 cc. was sufficient for adequate filling in the 7-month-old child, and 15 to 18 cc. was used in the older children. In the adults, as much as 30 cc. was injected but the average was approximately 20 cc. The dye was injected as rapidly as possible to produce a "bolus" of Diodrast travelling refluxly through the brachial into the subclavian artery. When it reached the origin of the vertebral artery the "bolus" was carried with the normal blood flow into the vertebral-basilar distribution.

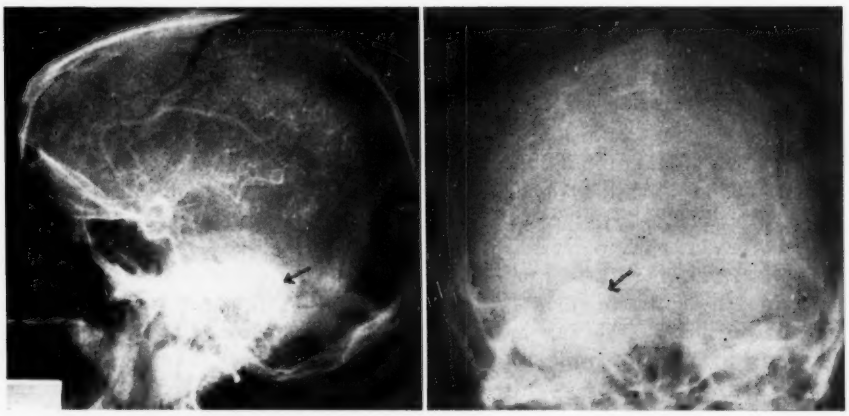

FIG. 1. Right retrograde brachial angiograms showing saccular venous angioma (arrow).

Three radiographic exposures were made with each injection. The first exposure was made just as the injection was completed: the others, at 1-sec. intervals thereafter. The anteroposterior exposure was made with the x-ray tube inclined 35° above a horizontal plane through the skull.

The needle puncture defect in the artery in children was closed with a single arterial suture of 5–0 silk; in adults the arterial suture was not necessary because pressure for a few minutes with a sponge resulted in adequate closure of the needle hole. The incision was closed with interrupted silk sutures and the arm was wrapped with an elastic bandage to prevent later formation of a hematoma.

With this technique both the vertebral and carotid systems are usually filled.

CASE EXAMPLES

Case 1 (#849327). D.H. was a 65-year-old male who had a sudden onset of a left hemiplegia 5 months before admission to hospital. Persistent vomiting began 1 month before admission. There was a spastic left hemiplegia and paresis of the right

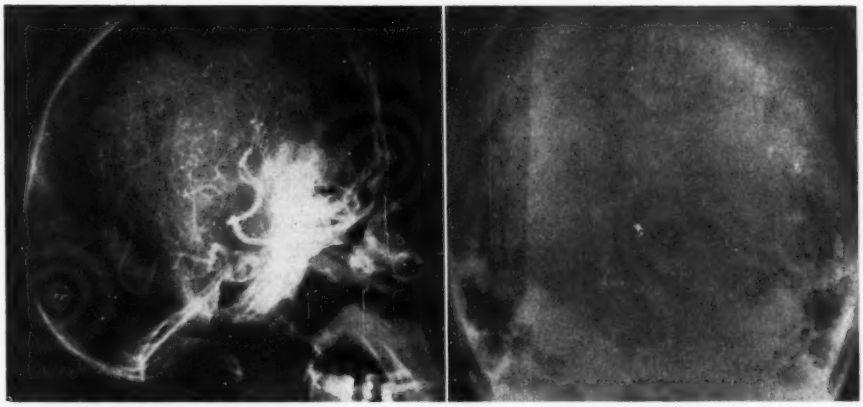

FIG. 2. Right retrograde brachial angiograms demonstrating vertebral and internal carotid circulation.

VI nerve. Roentgenograms of the skull revealed a shift of the pineal calcifications 5 mm. to the left. Brachial reflux angiogram (Fig. 1) revealed a 2.5 cm. saccular lesion communicating with the basilar vein. This was interpreted as a venous angioma, inaccessible to surgical excision. The patient expired 2 years later of a subarachnoid hemorrhage.

Case 2 (#858366). M.H., a 56-year-old housewife, had a spontaneous subarachnoid hemorrhage 24 hours before admission. Examination revealed nuchal rigidity

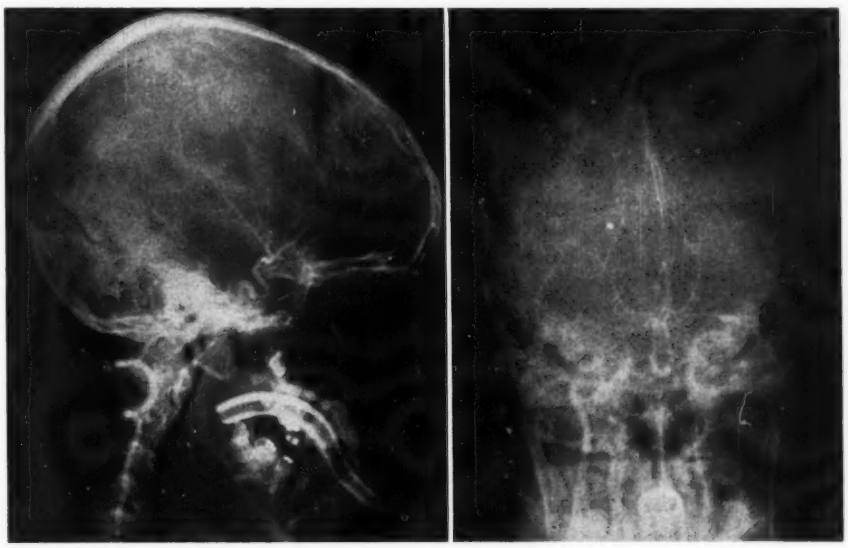

FIG. 3. Right retrograde brachial angiograms demonstrating vertebral and carotid circulation.

and blood in the lumbar cerebrospinal fluid. Bilateral carotid angiograms revealed no abnormality. Brachial reflux angiogram (Fig. 2) visualized the vertebral system without demonstrating a lesion. She was discharged from the hospital with a diagnosis of spontaneous subarachnoid hemorrhage, etiology undetermined.

Case 3 (#874566). Y.S., a 12-year-old girl, had a spastic left hemiparesis that developed 2 years prior to admission. Severe headaches and occasional vomiting began 3 months before admission. Examination revealed a left hemiparesis, hemiathetosis and bilateral papilledema. Brachial reflux angiogram (Fig. 3) showed a shift of the anterior cerebral artery to the left. On the lateral projection displacement of the same vessel indicated hydrocephalus. No definite vascular pattern of a tumor could be seen. A ventriculogram revealed evidence of a tumor of the right thalamus. At operation, a cystic astrocytoma was found.

FIG. 4. Right retrograde brachial angiograms. Left common carotid artery compressed in the neck. Note filling of both internal carotid systems and the vertebral circulation.

Case 4 (#877346). M.B., an 11-year-old girl, had three episodes of focal seizures beginning in the left foot and leg. Neurological examination revealed no abnormality. Brachial reflux angiogram (Fig. 4) visualized the vertebral and carotid system on the right. On the second injection for anteroposterior views, with compression of the left carotid in the neck, bilateral filling of the carotid systems occurred in addition to the vertebral-basilar circulation. No abnormality was noted. She was discharged to return later for a pneumoencephalogram.

COMPLICATIONS

The only complication observed in this small series of 16 patients was tingling in the fingers in one patient. It was felt that this was caused by the use of more than the usual traction of the nerves to expose the artery. The

tingling ceased upon relaxing the traction. No neurological deficit could be found after the procedure was completed. No attempts have been made to inject the brachial artery percutaneously because it was felt that the danger of trauma to the median and ulnar nerves exceeded the disadvantages of a minor incision.

The possibility of spasm occurring in the brachial artery has been considered but was not seen in any of these cases. Radner[4] and Hauge[1] reported that this complication usually occurs when the catheter reaches the subclavian artery. Radner[4] advised that a 2.5 per cent solution of papaverine hydrochloride be administered 1 hour before injection to prevent spasm from occurring. In none of our cases was medication employed to prevent spasm since the needle was introduced only 1–2 cm. into the brachial artery.

The site of the exposure was selected with the possibility that thrombosis might occur. Since the exposure is distal to the major collateral arteries the circulation should be entirely adequate if thrombosis occurs. In all of our cases radial arterial pulsations were easily palpated following removal of the needle.

The use of a stylet obviates the need for continuous saline in the interval between injections. The volume of saline needed to maintain patency of the lumen of the needle might overload an infant's circulation.

No patient exhibited any reaction from the injection of Diodrast other than the usual flushing sensation similar to that seen during carotid angiography. One adult received a total of 175 cc. of 35 per cent Diodrast in five injections in an attempt to obtain adequate filling.

DISCUSSION

This procedure has proven quite successful in children. In addition to the vertebral filling there has been excellent visualization of the ipsilateral internal carotid circulation in all the children except one. On the anteroposterior projection, contralateral compression of the carotid artery in the neck improves the ipsilateral filling of the carotid system and may lead to adequate visualization of the distribution of both internal carotid arteries (Fig. 4). The filling of the right internal carotid circulation has been sufficient with this technique to justify its use for investigating lesions in younger children in whom percutaneous puncture might be difficult. However, carotid arterial filling cannot be expected when the left brachial artery is injected because the contrast medium would need to travel through the left subclavian artery and then in a retrograde direction in the aorta to reach the origin of the common carotid artery.

The results with this technique in adults have not been encouraging. The adult arteries are proportionately larger and consequently the greater quantity of blood dilutes the "bolus" of contrast medium. In order to overcome this dilution factor it was necessary to inject excessively large amounts of dye. The possible risk with the use of such large dosages makes this technique in adults less feasible than percutaneous vertebral artery puncture.

CONCLUSION

A technique for performing vertebral angiography has been described. It is recommended for use in children in whom it is desired to investigate the vertebral or right internal carotid circulation. It is not recommended for use in adults except when other attempts at vertebral angiography by the usual methods have failed.

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HEREDITARY COMBINED NEURINOMAS AND MENINGIOMAS

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VON RECKLINGHAUSEN'S disease, in the form of multiple neurofibromatosis affecting not only peripheral nerves but cranial and spinal nerve roots, has frequently been demonstrated to show marked hereditary tendencies. Even when this disease, unaccompanied by widespread neurinomatosis, manifests itself only in the form of bilateral acoustic nerve tumors, it has been shown to be hereditary. In the remarkable report of a family by Gardner and Frazier,[2] they traced the bilateral deafness as a true Mendelian dominant through five generations comprising 217 persons.

Cushing and Eisenhardt,[1] in their classical monograph on meningiomas, have brought together in a most exhaustive manner the reports from the literature of cases of multiple meningiomas, and combined neurinomas and meningiomas occurring in the same patient. It is, indeed, with one of the two examples presented by these authors in Chapter V of their book, entitled "Combined Neurinomas and Meningiomas," that we are dealing in the present report. Stanley G., the subject of Case I, Serial No. 311, in Chapter V of their monograph, was referred to the care of one of us by Dr. Cushing upon his retirement from active neurosurgical practice. The present paper is a continuation of the remarkable history of this patient, and that of his only child. For the sake of completeness, we are taking the liberty of repeating the earlier history of Stanley G., as reported by Cushing and Eisenhardt, and will then report further upon our own experiences with this unique father and daughter.

CASE REPORTS

"Case I. *Serial No. 311. Multiple psammomeningiomas involving cerebral dura, roentgenologically verified, and symptomatically associated with bilateral auditory disturbances presumably due to bilateral acoustic neurinomas. Suboccipital exploration with surgical verification of multiple tumors supposedly meningiomas in the right lateral recess adjacent to the porus acusticus. Recovery with fairly stationary condition after 5 years.*

"*July 17, 1932.* Admission of Stanley G., a Jewish medical student, 24 years of age, complaining of headaches and impaired hearing. The family history revealed a tendency toward familial psychoneurosis.

"*Past History.*—For a period of at least ten or twelve years the patient had

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† Walter Reed Army Hospital, Washington 12, D. C.

noticed a tendency to flushing and sweating of the right half of the body. A large lipoma was removed from the suboccipital region in *1922*. For five years, progressive impairment of hearing and for four years inequality of pupils had been observed. An advancing right-sided deafness with tinnitus had been present for two years and tinnitus referred to the left ear for a year and a half. Some impairment of vision also had been noted for two and a half years; and latterly he had experienced colour hallucinations on stooping. Unsteadiness in gait with a tendency to deviate to the right and backward with attacks of weakness and trembling had been of recent occurrence. Despite a negative Wassermann he had received a thorough antiluetic treatment.

"*Physical Examination.*—This disclosed: (1) a left relative hyposmia; (2) bilateral choked discs of 2 diopters; (3) anisocoria, right pupil wider than left; (4) nystagmus poorly sustained; (5) hypaesthesia of the right trigeminal skin field; (6) slight palsy of the right face; (7) impaired hearing in the right ear with sluggish vestibular response to caloric test (normal reaction on left); (8) a definite right-sided tendency to flushing and hyperhidrosis; (9) positive Romberg with falling backward and to the right; (10) slight sensory loss over right arm and leg; (11) increased deep reflexes on the left; (12) a smooth cranial hyperostosis palpable in the left frontal bone with enlargement of the temporal artery.

"X-ray films showed unmistakable evidence of multiple psammomatous meningiomas bilaterally situated and involving the cranial vault. They were chiefly parasagittal in situation, more particularly on the left side in correspondence with the palpable hyperostosis.

"*Diagnosis.*—It was clinically apparent that there was a growth in the right cerebellopontile angle, probably an acoustic tumor associated with bilateral parasagittal meningiomas over the cerebrum. Ventriculograms disclosed moderate dilatation of the lateral ventricles with slight depression of both frontal horns.

"*Operation.—July 25, 1932.* Novocaine. Suboccipital exploration. The usual bilateral exploration was made. A tense cerebellum was disclosed and on exploring the right recess normal-appearing 9th, 10th, and 11th, and 7th and 8th nerves were seen. Closely adjacent to the porus acusticus were two tiny tumors supposedly meningiomas about 1 cm. in diameter, apparently attached to the dura. The first proved on supravital examination to be a psammoma; the other was not examined. It was thought to be another meningioma (*cf.* infra). The operation was then concluded.

"*Subsequent Notes.*—Convalescence from this procedure was protracted and stormy due to extreme vertigo and vomiting on the slightest change of position. In view of these distressing symptoms it was thought best to postpone an attack upon the meningiomas involving the forebrain. On *August 20, 1932* he was discharged.

"In spite of his handicap and increasing deafness, he courageously resumed his medical course and succeeded in graduating in *June 1936*. He meanwhile had married and become a parent. On *October 17, 1936* a thorough otological examination by Dr. Page Northington showed a total loss of vestibular response and air conduction on the right and partial loss in the left ear. In a report *April 12, 1937*, he stated that he was busy as an interne; that his general health was excellent, his mind active, his domestic life happy, and he was not in the least sorry for himself.

"*Histological Note.*—The first and smaller of the two tumors as stated was a psammomeningioma. When we came to cut the other, it proved to be a transitional type of neurinoma showing in addition to pali-

sading parallel fibres of fine reticulin, numerous whorls and in one area a few ganglion cells—in short, much the same appearance shown in the Crowe tumor. Dr. Bailey has shown us sections from two unpublished examples of central neurinomatosis with meningiomatosis, certain of the tumors being transitional lesions resembling the minute Crowe neurinoma and that observed in this last case—lesions, in other words, difficult to differentiate in that they have certain architectural features pertaining to both neurinomas and meningiomas."

Subsequent History. He was followed by one of us until 1940 when his hearing disappeared completely, and visual acuity, because of chronic papilledema with associated secondary atrophy, was reduced to a point where the patient, for all intents and purposes, was both deaf and blind. The patient's father, who was also a physician, then urged us to operate on the parasagittal tumors. By this time he had an enormous hyperostosis, and a surgical attack was made on it by a two-stage procedure at which a large extracranial soft-tissue tumor mass was removed, then a large part of the calvarium, including multiple small and large meningiomas. During the latter part of the procedure a number of veins leading into the superior longitudinal sinus were injured. Bleeding, however, was immediately controlled by the placement of a large chunk of muscle which was held in readiness for just such an occasion, and the wound was closed.

Following this operation, apparently because of the injury to these veins, the patient showed bilateral signs suggestive of superior longitudinal sinus thrombosis. Upon his discharge from the hospital a small draining sinus developed in his wound, from which bits of wax were discharged. After a number of these pieces came away, the wound healed. He made a slow recovery from his operation and in the course of about six months had a restoration of vision sufficient so that he was able to read newspapers with the aid of telescopic lenses, and while his life consisted largely of a wheel-chair existence, he enjoyed his family and friends, and kept in remarkably close touch with world and medical events. However, about a year after the operation he had a sudden unexplained fever with temperature rising to 105°, and a correspondingly high leucocyte count. After a few days he went into coma and died on February 24, 1942. Unfortunately no postmortem examination was permitted.

Case II. *Solitary spinal cord meningioma removed in 1949, when patient was 15 years old, followed, five years later, by bilateral auditory disturbances presumably caused by bilateral acoustic neurinoma. Suboccipital exploration with surgical verification of multiple tumors in both lateral recesses, histologically diagnosed as meningiomas.*

July 6, 1949. Admission of daughter of Stanley G. with chief complaint of numbness and instability of both legs and feet.

History. This was a 15-year-old schoolgirl whose past history was not remarkable except that her basal metabolic rate was somewhat low, and she had been taking thyroid extract to correct this. In May, 1949, about two months before admission to the hospital, she began to be aware of numbness in both feet, gradually ascending up to her knees and involving the left side more than the right. This was associated with weakness of her legs and instability of gait. Her left knee began to give way suddenly and unexpectedly while walking, and she had several rather severe falls. Except for a feeling of numbness between her shoulder blades, there was no pain and no history of tingling, paresthesias, or incontinence, although transient episodes of urinary urgency had occurred in the two weeks prior to admission.

Physical Examination. The patient was a healthy-looking, pleasant, intelligent,

rather plump little girl. Her vital signs and general physical findings were normal. Neurologically the examination disclosed a paraparesis, with hyperactive knee and ankle jerks, and bilateral positive Babinski signs. On sensory examination, a sensory level below which pain, temperature and touch were diminished, was noted at about the 10th thoracic dermatome. Position sense was intact in her toes, but vibratory sense was lost up to the pelvis. No percussion tenderness of her spine was noted.

Laboratory Data. Routine studies of the blood and urine were negative. Roentgenogram of the thoracic spine was normal. Measurements between the pedicles were normal on the roentgenogram. Lumbar puncture revealed a partial block,

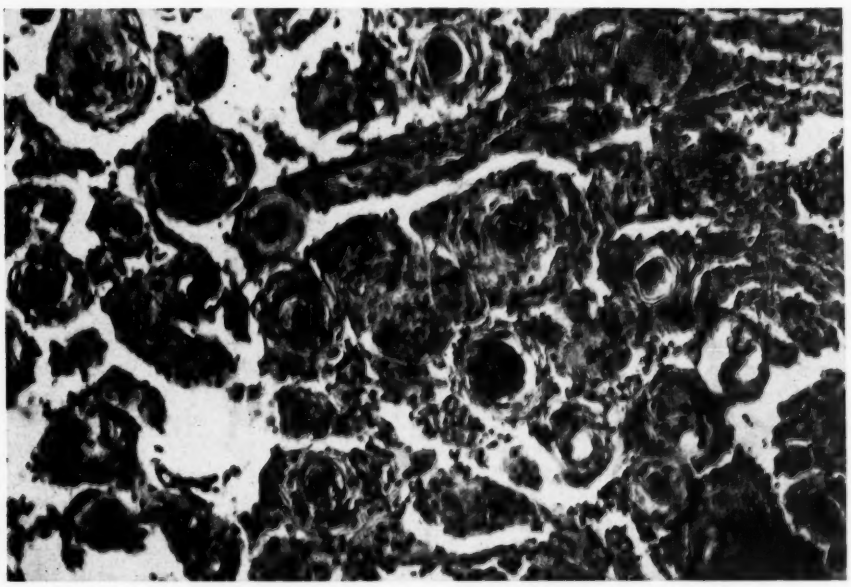

FIG. 1. Case II. Low-power photomicrograph of the spinal cord tumor, showing multiple psammoma bodies and whorls typical of meningioma.

and myelograms with 6 cc. of pantopaque proved the block to be at the level of the 5th thoracic vertebra. The inferior margin of the defect showed a double curve "like a mitten and thumb."

Operation, July 8, 1949. A complete laminectomy of thoracic vertebrae 4, 5 and 6 was performed and a typical, well encapsulated, intradural, extramedullary tumor was found adherent to the dura mater and indenting the spinal cord, chiefly from the right side. The tumor, together with the dura mater to which it was attached, was removed in one piece. The specimen measured 2.5 by 1.5 by 1 cm.

Histological Note. On microscopic examination the picture was that of a classical psammomatous meningioma, with typical multiple whorls, many of which contained lime salt deposits (Fig. 1).

Subsequent History. Immediately following operation the patient showed an increase in her neurological signs in the form of increased weakness, disturbances of

bladder, and sensory changes. This led us, after 24 hours, to reexplore the wound for fear that a postoperative clot may have occurred. The suture line in the dura mater was carefully opened and the cord was inspected. The arachnoid seemed to be free. The cord was not edematous, nor did it seem soft and mushy. There appeared to be no evidence of thromboses of the vessels on the surface of the cord, so that we were left without explanation as to the reason for the increased signs. For purposes of decompression the dura mater was left open on this occasion.

Within the next week the patient made an excellent recovery, and except for slightly exaggerated reflexes and persistent Babinski signs, she walked out of the hospital well, for all intents and purposes.

Although she occasionally wrote one of us, from which we learned that she

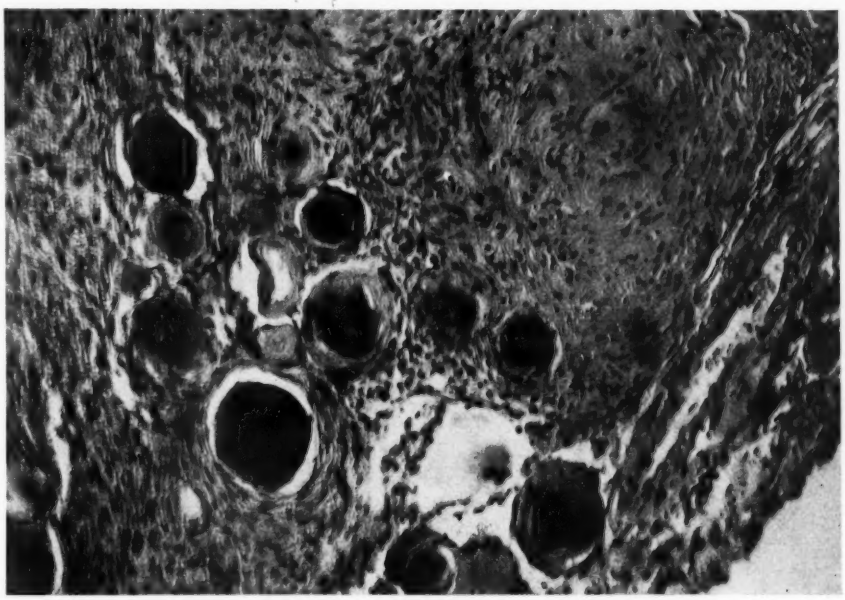

FIG. 2. Case II. Low-power photomicrograph of the biopsy specimen removed from the posterior fossa at operation.

completed high school, went on to college and eventually married a young soldier, she was not seen again until July 14, 1954, when she was complaining of the symptoms that led to her second operation. Since the young couple was extremely limited in their financial means, and since as a soldier's wife she was entitled to service at the Walter Reed Army Hospital in Washington, D.C., where she had already been seen once, it was there that the following developments took place.

July 16, 1954. Admission to Walter Reed Army Hospital.

History. The patient's present illness began about ten months prior to her admission, in August, 1953, when she noted an echoing of her own voice in her head and some bilateral tinnitus. The symptoms gradually increased in severity but never became disabling. Three months prior to her admission she had measles, and

her recovery was accompanied by an onset of staggering gait, marked increase in deafness and tinnitus, and numbness of the left side of the face. She also suffered, for the first time, from intermittent headache chiefly in the left occiput and left mastoid areas. Generalized headache occurred when she turned her head suddenly in either direction. Diplopia developed in all but the far left lateral gaze. Since the onset all these symptoms progressively increased in severity.

Physical Examination. The patient walked with a lurching gait, constantly veering to the right. She was very deaf to air conduction in both ears, but seemed to

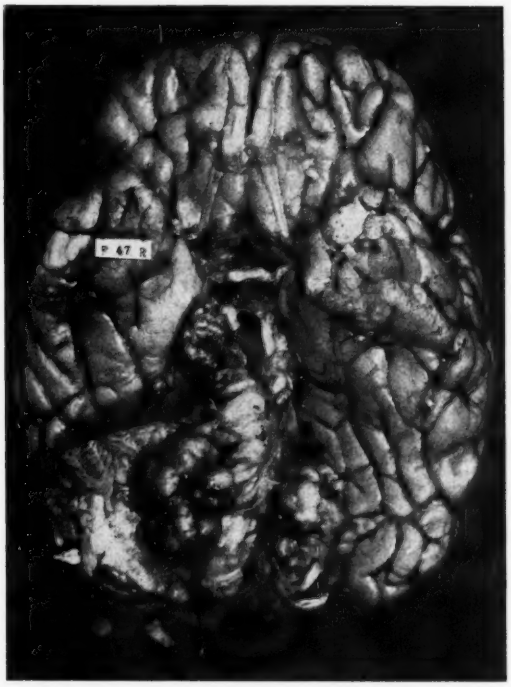

FIG. 3. Case II. Basilar view of brain post mortem showing the two cerebellopontine angle tumors.

perceive higher frequency sounds better than the lower ones. Caloric testing revealed no response to ice-water circulation on either side. There was no evidence of neurofibromata or stigmata of von Recklinghausen's disease anywhere on the skin of her body. Slight diminution in the sense of smell was detected in the right nostril. Bilateral long-standing papilledema was evident without accompanying hemorrhages or exudates. Visual fields were grossly normal. A partial right 6th nerve paresis was present. The left corneal reflex was diminished as compared to the right. Hypalgesia and hypesthesia were present over the entire distribution of both trigeminal nerves, more marked on the left. No motor trigeminal weakness was demonstrable. A moderate peripheral 7th nerve weakness bilaterally was also seen, more marked on the right. Gross motor weakness of the left upper extremity and

hemihypalgesia involving the entire left side of the body were detected. Bilateral Babinski signs were demonstrated, which have been present since the removal of her spinal cord tumor. There was no ankle clonus, and Hoffmann sign was negative bilaterally. Coarse nystagmus on left lateral gaze and fine nystagmus on upward gaze were seen. Deep tendon reflexes were within normal limits, but were more active on the left than on the right side. Abdominal reflexes were absent on the right. Past-pointing and rebound phenomena were demonstrable bilaterally, more marked on the left. She seemed to fall, however, to the right on Romberg testing.

Laboratory Data. Routine laboratory tests were negative. Roentgenograms of her thoracic spine showed the old laminectomy of thoracic 2 to 5, with residual contrast medium within the skull, apparently in the basilar cisterns to the left of the sella

FIG. 4. Case II. Postmortem specimen of cerebral and cerebellar dura mater, showing multiple, adherent, small, discrete and confluent meningiomas.

turcica. Roentgenograms of the skull revealed asymmetry of the petrous bones, and the right side showed some erosion at the tip. Both internal auditory meatuses appeared to be enlarged, that on the right being larger than the one on the left.

Operation, August 6, 1954. A bilateral suboccipital craniectomy was performed, and the left ventricle was punctured through a separate incision and burr hole. This did not entirely relieve the tension under which the dura mater over the cerebellum was found, but evacuation of the cisterna magna made the opening of this membrane safe. The cerebellar hemispheres did not appear to be under pressure when exposed. They were of equal size and the vascular markings over them, the vermis and the tonsils were entirely normal. There was no herniation of the tonsils into the foramen magnum. The posterior third of the rim of the foramen magnum was removed. The left cerebellopontine angle was inspected first, and a large looping posterior inferior cerebellar artery was encountered. Just anterior to this a mass of grayish-white

tumor tissue, partly adherent to the dura mater in the entire area, spreading over the region of the sigmoid sinus and going far up into the angle, was seen. Four or five pea-sized nodules of this firm, pasty, matrix-like tumor mass were removed for biopsy, but the mass of tumor itself was considered surgically unremovable since it appeared to be surrounding not only the cranial nerves from the 5th nerve posteriorly, but also many large blood vessels. The biopsy showed typical meningioma (Fig. 2).

The right cerebellopontine angle was then inspected. The angle itself was fairly clean of tumor, but down toward the 9th, 10th, 11th, and 12th nerves there was a nodule of tumor intimately associated with a loop of the posterior inferior cerebellar

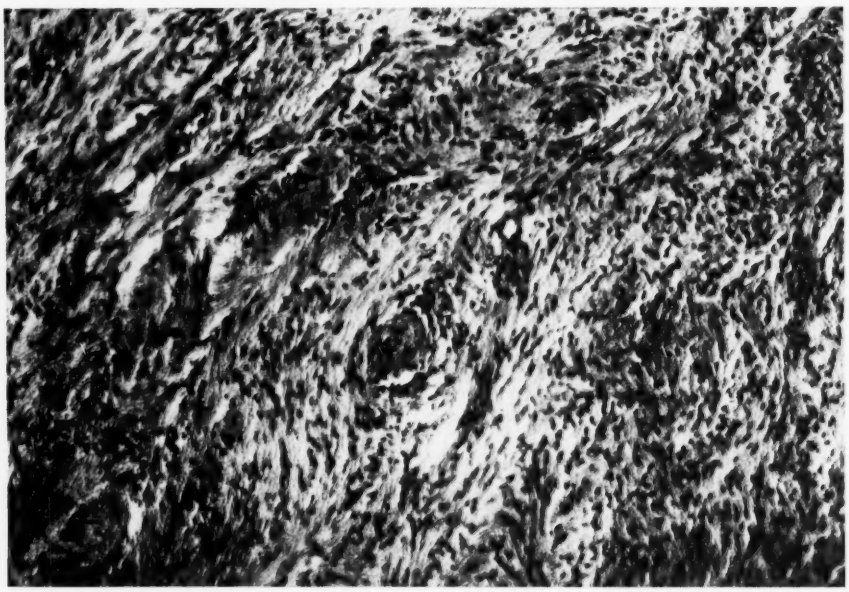

FIG. 5. Case II. Low-power photomicrograph of left cerebellopontine angle tumor, typical of neurinoma but showing a whorl formation in the center of the field.

artery. Since the tumor obviously was on both sides, and probably crossed the midline anterior to the pons in a solid sheet, the operator felt it was foolish to persist, and left the patient with a simple decompression and biopsy.

Subsequent Course. The patient made a very uneventful postoperative recovery, but by the time of her discharge from the hospital on August 20, 1954, she had shown no evidence of improvement in any of her cranial nerve deficits, although some slight subjective improvement in the strength of her left hand grip was noted.

About a month after her discharge the patient was found dead in a hotel room under somewhat suspicious circumstances, and a postmortem examination was made by the Medical Examiner.

Autopsy. The general necropsy was without interest for this presentation.

The brain showed two fairly large-sized tumors, one in each cerebellopontine

angle. The tumors were encapsulated, lobulated, and measured about 5 by 4 by 3 cm. each (Fig. 3). In addition there were multiple, almost countless, small, discrete as well as confluent tumor nodules on the undersurface of the dura mater, along the falx and superior longitudinal sinus, the lateral sinuses, and the upper and lower surfaces of the tentorium (Fig. 4). These small lesions, histologically, proved to be typical meningiomas. The two tumors in the cerebellopontine angles, however, were quite classical examples of neurinomas with palisading and parallel fibres of

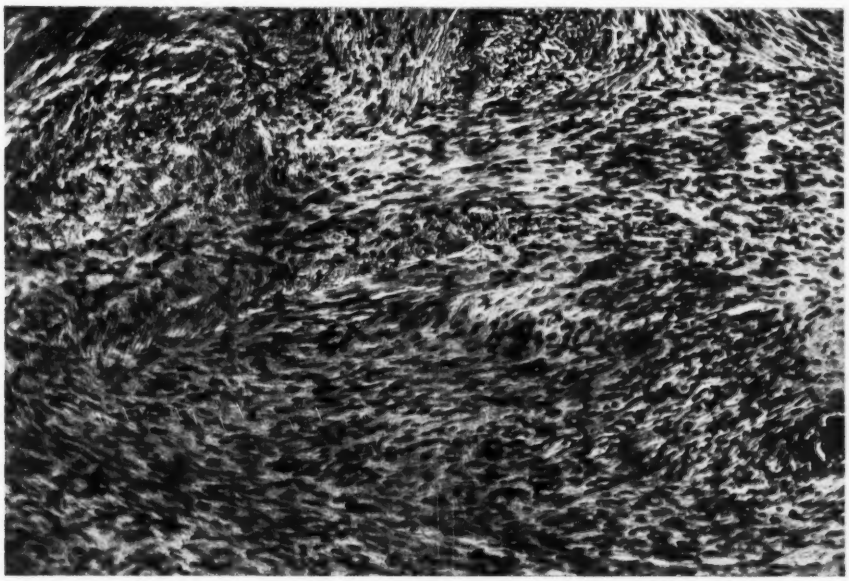

FIG. 6. Case II. Similar view of right cerebellopontine angle neurinoma.

fine reticulin, but showed, in areas, whorl formation, and in several places ganglion cells (Figs. 5 and 6).

DISCUSSION

The purpose of this report was to place on record these two remarkable cases. The many implications, including the nature and the multiplicity of these lesions, their relationship to von Recklinghausen's disease, the interrelationship between meningiomas and neurinomas, and even the light this may throw on the germ layer origin of the meninges, are so completely covered in the monograph of Cushing and Eisenhardt that it would be repetitive to present them here again.

SUMMARY

Two cases of bilateral cerebellopontine neurinomas combined with meningiomatosis are reported, occurring in a father and daughter. In both

patients bilateral deafness was present, and in each bilateral acoustic neurinomas were suspected. In the father a small tumor nodule in one angle was found which, in addition to typical whorl formations, also showed areas of palisading suggesting a mixed type of tumor. The rest of his many small and large intracranial neoplasms were typical meningiomas. In the daughter a meningioma from the thoracic spinal canal was removed at the age of 15, and at 20 multiple posterior fossa meningiomas were verified at operation. At postmortem examination the undersurface of the dura mater bilaterally was studded with discrete as well as confluent almost countless meningiomas, in addition to which two larger tumors, one in each cerebellopontine angle, were found which histologically were neurinomas, but contained whorls in areas suggestive of mixed neurinoma and meningioma in the same tumor, similar to the angle tumor reported in the case of her father.

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STUDIES ON THE ROLE OF THE CHOROID PLEXUS IN TRACER EXCHANGES BETWEEN BLOOD AND CEREBROSPINAL FLUID*

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ALTHOUGH the choroid plexuses of the cerebral ventricles are generally accepted as the site of cerebrospinal fluid formation, the problem of the rate of formation of cerebrospinal fluid and blood-cerebrospinal fluid tracer exchange before and after choroid plexectomy has never been directly attacked. Therefore, it is pertinent to the general problem of cerebrospinal fluid formation and circulation to examine the effect of choroid plexectomy on various ionic and molecular tracer exchanges between blood and the cerebrospinal fluid. This paper presents the results of an investigation of these exchanges in dogs and some neurosurgical patients undergoing choroid plexectomy.

MATERIALS AND METHODS

All of the animal experiments have been carried out on mongrel dogs 15 kg. to 20 kg. in weight. The patients studied were hydrocephalic infants who were having choroid plexectomies carried out as a therapeutic measure.

The tracers used were heavy water (D_2O), Na^{24}, K^{42}, and I^{131} tagged human serum albumin (RISA). These tracers were injected intravenously and samples of arterial blood, ventricular, cisternal and lumbar cerebrospinal fluid were taken at intervals for determination of the tracer concentration. All cerebrospinal fluid samples were 0.2 ml. or less. All tracer values are calculated as specific activity. If A* is a tracer of substance A, then the specific activity of any sample would be calculated as follows:

$$\text{Specific activity} = \frac{\text{amount of tracer A* present in the sample}}{\text{total amount of A in the sample}}$$

The results calculated in this way show the relative exchange regardless of the absolute concentration of the substance considered and it allows comparison of the exchanges of various substances. Comparison of experiments with the same tracer is sometimes difficult because of variations in the amount of tracer injected. This difficulty has been resolved by assigning the value 100 to the tracer specific activity of the serum 3 hours after injection of the tracer. Observed specific activity values are then recalculated by the following formula:

* This investigation was supported by a research grant (B-157 C2) from the National Institute of Neurological Diseases and Blindness, of the National Institutes of Health, U. S. Public Health Service.

$$\text{Relative specific activity of a sample} = \frac{\text{observed specific activity of samples} \times 100}{\text{specific activity of serum at 3 hours}}$$

Hydrocephalus was produced in the dogs by cisternal injection of 750 mg. kaolin. The choroid plexectomies and plugging of the foramen of Monro were carried out through occipitoparietal incisions in the cerebral cortex. Gelfoam soaked in kaolin was used to plug the foramen of Monro. Dye was injected intraventricularly to test for possible communication between the ventricles, and through choroidal fissures to the subarachnoid space. Postmortem examinations were carried out on all dogs.

RESULTS

This paper is concerned with the choroid plexus, so only the ventricular appearance of the tracers will be considered, although it is known that they do appear independently in other portions of the cerebrospinal fluid.[4,6,16–19]

The appearance curves of various tracers in the ventricular cerebrospinal fluid of the normal dog are shown in Fig. 1. These curves are similar to those

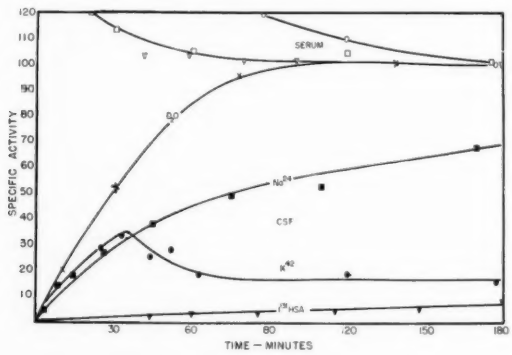

FIG. 1. The appearance of D_2O, Na^{24}, K^{42}, and I^{131} tagged human serum albumin in the cerebrospinal fluid of the lateral cerebral ventricle of a normal dog. The equilibrium serum concentration of each tracer was taken as 100 in computing the specific activities.

seen in humans. It is at once apparent that each tracer exchanges at a different rate and probably independently. Water exchanges much faster than any of the other substances and protein more slowly. Sodium and potassium fall between these two, but they are not alike. Sodium exchange proceeds slowly and steadily with the specific activity of the cerebrospinal fluid reaching equilibrium in a few hours. Potassium is, on the other hand, quite different. The K^{42} exchange between blood and cerebrospinal fluid takes place quite rapidly until the specific activity of the cerebrospinal fluid is about 15–20 per cent that of the blood and then the specific activity of the cerebrospinal fluid approaches that of the blood very slowly. These observations of various appearance curves for various substances have been confirmed by several others.[6,9,16–19] It is not within the scope of this paper to discuss the reasons for the differences between these various

appearance curves, but it is obvious that no one of these tracers can be taken to represent cerebrospinal fluid as a whole. Each tracer must be considered by itself and accumulation of cerebrospinal fluid treated as a separate phenomenon.

Water Exchange. Water tracer (D_2O) exchange between blood and cerebrospinal fluid has been measured before and after choroid plexectomy in two lnfants with obstructive hydrocephalus. There was no change in the D_2O exchange after removing the choroid plexuses of both lateral ventricles. One of the patients was found to have a small adenoma of one choroid plexus. The studies on this patient are shown in Fig. 2.

The apparent slowness of the D_2O exchange curves in this patient as compared to the normal D_2O exchange curve is the result of his large ven-

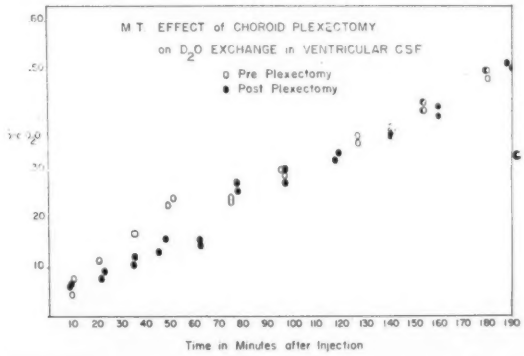
M.T. EFFECT of CHOROID PLEXECTOMY
on D_2O EXCHANGE in VENTRICULAR CSF
o Pre Plexectomy
• Post Plexectomy
60
50
20
10
10 20 30 40 50 60 70 80 90 100 110 120 130 140 150 160 170 180 190
Time in Minutes after Injection

FIG. 2. Heavy water (D_2O) exchange between blood and ventricular cerebrospinal fluid of an infant with obstructive hydrocephalus. Samples were taken from both ventricles in studies done before and after bilateral choroid plexectomy.

tricles and not any basic change in the rate of water exchange. This concept has been discussed in another paper.[2] Although there are one or two variations between the pre- and post-plexectomy points on this chart they are within the limits of experimental error and the two curves can be regarded as identical.

Sodium and Potassium Exchange. The sodium and potassium exchanges were studied in dogs before and after removal of the choroid plexuses of the lateral ventricles. Typical curves are shown in Fig. 3. These were repeated in four animals with the same result. The sodium exchange showed no variation after removal of the choroid plexuses of the lateral ventricles. The potassium exchange in some experiments was altered in its fast early phase. However, it was not possible to get enough samples of cerebrospinal fluid during this period to determine whether or not this alteration of the fast portion of the exchange curve occurred as the result of the choroid plexectomy. There was no suggestion of a difference in the slow phase of exchange seen after 35 minutes.

These experiments did not entirely eliminate the choroid plexus because

the lateral ventricles communicated with the III and IV ventricles where the choroid plexuses remained. Complete isolation of the cerebrospinal fluid from choroid plexus was obtained in hydrocephalic dogs by doing a unilateral choroid plexectomy and plugging the foramen of Monro on the same side. In these animals simultaneous studies were made of the appearance of sodium and potassium in the fluid of each ventricle. The exchange of the tracers between blood and the ventricular cerebrospinal fluid was a bit faster in the ventricle without the choroid plexus, as might be expected

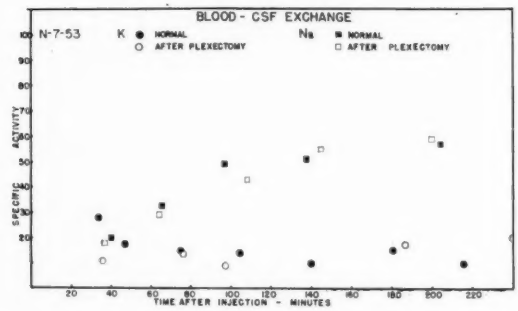

FIG. 3. Na^{24}, K^{42} exchange between blood and ventricular cerebrospinal fluid in dogs before and after choroid plexectomy.

because of its slightly smaller size. These studies were repeated four times with the same result. Typical studies on two dogs are shown in Fig. 4.

Tagged Albumin Exchange. I^{131} tagged albumin (RISA) exchange between blood and ventricular cerebrospinal fluid in two dogs is shown in Fig. 5. There is some apparent scatter of the points on the pre- and post-plexectomy curves of both animals. The post-plexectomy points of one animal (N-154-52), shown as squares, tended to be slightly higher than the pre-plexectomy points, while the opposite was true of the other animal (N-21-53), shown as circles. However, these variations are within the combined errors of the protein determinations and radio-active determinations of these very small samples (0.1 ml. samples of cerebrospinal fluid contained only about 0.02 mg. of protein of which only a small amount was tracer). The over-all trend of the RISA appearance curve was not significantly altered by the removal of the choroid plexuses.

DISCUSSION

The experiments in which only the choroid plexuses of the lateral ventricles were removed are not conclusive because the choroid plexuses of the III and IV ventricles were still present. However, the results are highly suggestive of a lack of importance of the choroid plexus in the ionic and molecular exchanges considered. This conclusion is supported by the group of experiments in which the ventricles were isolated from each other and

one was without a choroid plexus. These experiments demonstrated Na^{24} and K^{42} exchange between blood and cerebrospinal fluid in the absence of the choroid plexuses as easily as when the choroid plexus is present. In addition, these experiments are supported by studies on the pulse pressure effects of the choroid plexuses and cerebrospinal fluid drainage experiments.[3–5]

Another hypothesis that is suggested is that the brain compensates for the loss of exchange by the choroid plexuses. This implies the assumption that certain rates of molecular exchange must be maintained. However, this idea not only is unsupported, but there is contrary evidence from the work of Bakay[1] on $PO4^{\equiv}$ exchange. His experiments suggest that $PO4^{\equiv}$ enters the cerebrospinal fluid from both the brain and choroid plexus. If the choroid plexuses were removed he found that $PO4^{\equiv}$ entered the cerebrospinal fluid more slowly than before, at a rate determined by the blood-brain $PO4^{\equiv}$

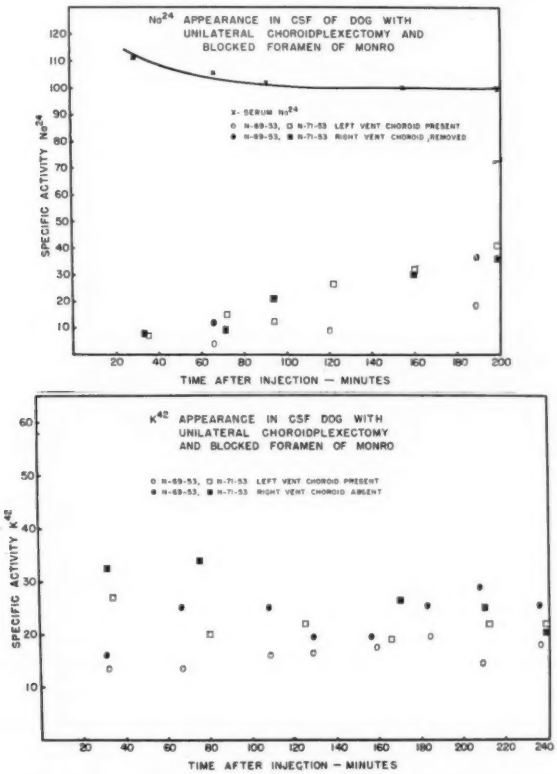
Na24 APPEARANCE IN CSF OF DOG WITH UNILATERAL CHOROIDPLEXECTOMY AND BLOCKED FORAMEN OF MONRO
x- SERUM Na24
o N-69-53, □ N-71-53 LEFT VENT CHOROID PRESENT
● N-69-53, ■ N-71-53 RIGHT VENT CHOROID REMOVED
SPECIFIC ACTIVITY Na24
TIME AFTER INJECTION — MINUTES
K42 APPEARANCE IN CSF DOG WITH UNILATERAL CHOROIDPLEXECTOMY AND BLOCKED FORAMEN OF MONRO
o N-69-53, □ N-71-53 LEFT VENT CHOROID PRESENT
● N-69-53, ■ N-71-53 RIGHT VENT CHOROID ABSENT
SPECIFIC ACTIVITY K42
TIME AFTER INJECTION — MINUTES

FIG. 4. K^{42} and Na^{24} exchange between blood and the cerebrospinal fluid of isolated cerebral ventricles, one of which had its choroid plexus removed and foramen of Monro plugged, and in the opposite ventricle the choroid plexus was intact.

exchange. These experiments not only show that the brain does not seem to compensate for loss of choroid plexus, but they also emphasize the different routes of exchanges for different substances.

The conclusion that the choroid plexus is not particularly important in the blood-cerebrospinal fluid exchange of D_2O, Na^{24}, RISA, and K^{42} (with the possible exception of the early phase) seems to be at variance with the popular concept that the choroid plexuses are the major site of formation of cerebrospinal fluid. However, these experiments are not at variance with the evidence that gave rise to this theory. It is not the purpose of this paper

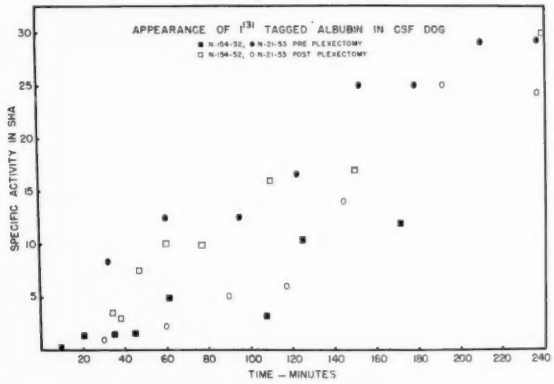

FIG. 5. I^{131} tagged albumin (RISA) exchange between blood and ventricular cerebrospinal fluid in dogs before and after choroid plexectomy.

to give a complete discussion of the theories of the formation of cerebrospinal fluid nor to formulate a new one, but it is of some interest to present a brief résumé of the primary evidence usually cited to support the theory that the choroid plexuses are the site of the formation of cerebrospinal fluid.

Clinical facts demonstrate that cerebrospinal fluid accumulates in the cerebral ventricles and if it cannot escape, hydrocephalus results. However, the clinical state of hydrocephalus does not give any evidence of the local site of formation of cerebrospinal fluid within the ventricular system.

Dr. Cushing's observation[7] of beads of fluid forming on the choroid plexus has often been quoted as positive proof of the plexus as the major site of formation, although it has been pointed out by many people, including Cushing,[7] that this observation was only suggestive and not conclusive. The appearance of fluid on the ventricular wall also has been reported[13] but it has not been repeated as often as Cushing's observation. During the course of the present experiments beads of fluid were more easily and more frequently seen to appear on the ventricular wall than on the choroid plexus.

Schaltenbrand and Putnam[15] observed fluorescein entering the cerebrospinal fluid from the choroid plexus after an intravenous injection. However, fluorescein is a special case and unless evidence is presented to show that it represents cerebrospinal fluid as a whole it can only be considered to

cover the movement of fluorescein from the blood into the cerebrospinal fluid.

Dandy[8] confirmed the clinical facts of hydrocephalus in dogs by showing that hydrocephalus would result if the subarachnoid spaces around the brain stem were occluded, if the aqueduct of Sylvius were occluded, and if the foramen of Monro were occluded the blocked ventricle would enlarge. He further demonstrated that if a unilateral choroid plexectomy and plugging of the foramen of Monro were carried out before plugging the aqueduct of Sylvius, unilateral hydrocephalus would result. The ventricle without the choroid plexus would not enlarge. He did not consider the fact that fluid was present in the ventricle without the choroid plexus, nor did he measure the rate of fluid accumulation in either ventricle. The pressure effects were not considered, although no instruments were available at that time to make proper measurements. Pressure effects have been recently considered in another paper.[3]

There have been many more papers written[14] on the formation of cerebrospinal fluid but since the publication of Dandy's paper in 1919, most investigators have assumed the production of cerebrospinal fluid by the choroid plexuses as a primary basis for the interpretation of their data. However, it should be pointed out that the concept of formation of cerebrospinal fluid by the choroid plexus was not universally accepted. It was vigorously opposed by Hassin[10,11,12] on pathological grounds. He put forward the theory that cerebrospinal fluid came from the brain and that the choroid plexus served to remove the cerebrospinal fluid or certain waste products in it. Wallace and Brodie[20] studied the distribution of bromide and iodide ions in the central nervous system and used these substances to identify the cerebrospinal fluid. They concluded the cerebrospinal fluid came from the intercellular fluid of the brain via the perivascular spaces.

All of the early investigators have assumed cerebrospinal fluid to move as a whole of fixed composition and that any one substance could be used to identify it as a whole. The variation of the ionic appearance curves shows this to be a false concept. Instead the cerebrospinal fluid should be considered as a mixture of many things passing into and out of it in accordance with the physiological characteristics of each constituent. The phenomenon of accumulation of cerebrospinal fluid is separate from any one substance and must be considered by itself.

Anatomical considerations as well as the surgical observation of fluid coming from the choroid plexus make it certain that some exchange of tracers between blood and cerebrospinal fluid does take place through the choroid plexus. However, the results of these experiments suggest that for the tracers considered the choroid plexus exchange is relatively small compared with the exchange between brain tissue and cerebrospinal fluid. The final assessment of the relative importance of the brain and the choroid plexus as the sites of accumulation of cerebrospinal fluid cannot be made from these data or experiments of this type as the ionic tracers do not represent the cerebrospinal fluid as a whole.

SUMMARY

1. The exchange of Na^{24}, K^{42}, D_2O (heavy water) and I^{131} tagged albumin between blood and cerebrospinal fluid of the cerebral ventricle has been studied before and after choroid plexectomy. Choroid plexectomy did not seem to affect these exchanges, except possibly the early fast phase of K^{42} exchange.

2. Some exchange of these tracers may occur between the blood and the cerebrospinal fluid through the choroid plexus, but it seems to be relatively small compared to the exchange occurring with brain.

3. Accumulation of cerebrospinal fluid should be considered as a special phenomenon separate from tracer ion exchanges.

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STUDIES OF TRIGEMINAL NERVE POTENTIALS*

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AMONG the numerous mechanisms proposed to explain the spontaneous and touch-induced pain of trigeminal neuralgia have been pressure upon the posterior root in the posterior fossa by vascular malformations and anomalies;[1,6] pressure on the posterior root by narrowing of the dural channel at the petrous tip;[10] arteriosclerotic changes in the Gasserian ganglion;[7] spastic vascular crises producing an acute anoxemic state of the nerve;[9] sensory epileptiform discharges or release of an efferent sensory inhibitory mechanism in the trigeminal system;[14] and "an electrical short circuit, so that benign stimuli of touch are deflected within the pain pathway" secondary to vascular disease or neurotropic virus.[2]

The present investigation was undertaken to determine the possible association of touch and pain fibers in the trigeminal system. Trigeminal nerve, ganglion, root, tract and nuclear potentials have been studied. Experiments were carried out on 16 cats under very light Surital anesthesia with Anectine and endotracheal artificial respiration. The head was immobilized and a portion of one cerebral hemisphere and cerebellum was removed, exposing the trigeminal system from the foramen rotundum to the obex of the medulla. Rectal temperatures were recorded.

Stimuli were delivered from a Grass stimulator, to subcutaneous filaments of the infra-orbital nerve after piercing the skin with two probe electrodes 1 cm. apart; to a dissected peripheral branch of the maxillary division, or to the 2nd division in the middle fossa. Unipolar shocks of 0.01 msec. duration were delivered at increasing voltages, evoking conducted responses from threshold to maximum.

The recording electrodes were paired insulated bipolar steel needles. The uninsulated tips were 60–80μ in diameter and separated by 0.5 to 1 mm. Such electrodes were inserted parallel to the long axis of the 2nd division, Gasserian ganglion, posterior root, the spinal tract and nucleus, and the ascending tract and main sensory nucleus of the trigeminal system, as shown in Fig. 1. The potentials derived from each pair were led through a push-pull amplifier to a conventional oscilloscope. All recording sites in the medulla were electrolyzed for histological identification.

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† Markle Scholar in Medical Science.

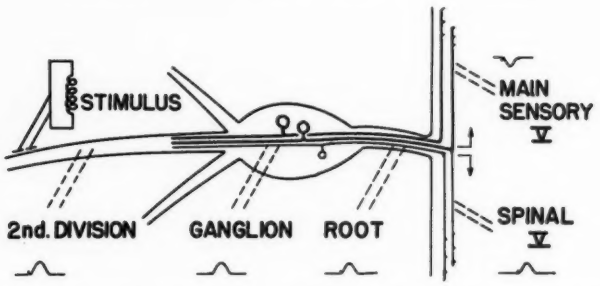

FIG. 1. A diagrammatic scheme of electrode placement for recording potentials from the trigeminal system following stimulation of the 2nd division.

RESULTS

Fig. 2 contrasts a typical response from the ganglion and root following stimulation of subcutaneous nerve filaments and the 2nd division in the middle fossa. Similar records may be obtained by dissecting a distal cutaneous twig of the maxillary nerve for stimulation, but the added time required for the dissection and the increased difficulty encountered in balancing the shock artifact, have led us to choose the subcutaneous skin probes for stimulation in this initial survey. The fast spike signaling afferent conduction of the impulse at the ganglion and root has a longer latent period following the shock artifact caused by subcutaneous stimulation than the afferent spike recorded in the ganglion and root following stimulation of the 2nd division in the middle fossa. The delayed potentials that follow the initial afferent spike response are not significantly different in the two records. As will be shown later, these delayed potentials are similar in many respects to the dorsal root reflex of spinal nerves.

In Fig. 3, characteristic responses in the 2nd division, Gasserian ganglion

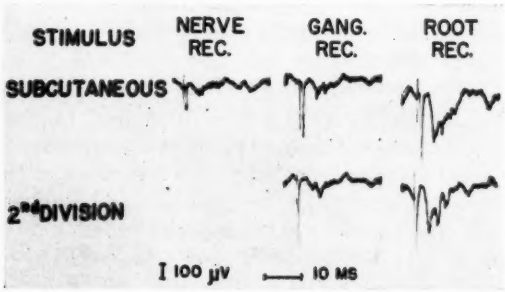

FIG. 2. Records from the 2nd division in the middle fossa, Gasserian ganglion and posterior root evoked by stimulation of subcutaneous nerve filaments in the 2nd division and stimulation of the 2nd division in the middle fossa. The clear separation of shock artifact from the afferent spike is evident following subcutaneous stimulation.

and posterior root are illustrated. The stimulus strength is given as a percentage of that required for maximum response. The delayed response in the 2nd division had a first crest (a) that was nearly maximal at 37 per cent with a delay of 3 msec. following the initial spike, and two or three succeeding crests (b, c) increasing in amplitude at higher threshold (50 and

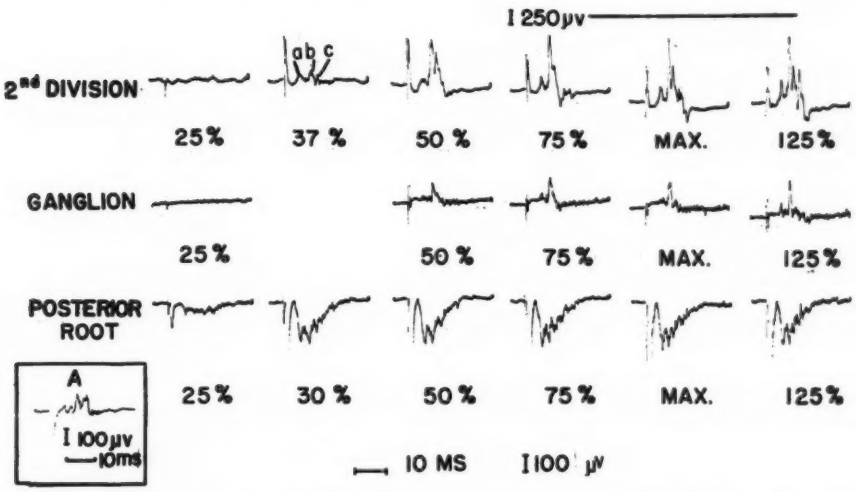

FIG. 3. The response is recorded in the trigeminal 2nd division, Gasserian ganglion, and posterior root. Stimuli increase from 25 per cent to 125 per cent of that required to produce a maximum response. The fast afferent spike follows the shock artifact after a brief interval. The three crests of the delayed response (a, b, c) appear after a further delay of 2.5 msec. in the nerve. Insert A illustrates the evoked delayed potential recorded in the 3rd division of the trigeminal nerve following stimulation of the 2nd division.

75 per cent maximum), indicating activation of axons of smaller diameter. The total duration of the response was 10–20 msec.

The response in the ganglion was usually of lower voltage than in the root but contained the same components.

In the posterior root the records showed (1) less delay at maximum between the afferent spike and the initial crest of the delayed response; (2) little difference in the threshold for all elements of the response and (3) higher voltage in the initial crest. In the posterior root, in addition to exteroceptive afferents from the 1st and 2nd divisions, there are also proprioceptive elements of the 3rd division and motor axons. The differences in the delayed potentials from the root in contrast to the 2nd division may be caused by this addition of large proprioceptive and motor fibers in the root. It is possible to show that an afferent stimulus evoked by subcutaneous stimuli to the 2nd division may be followed by a delayed discharge in the 3rd division as well as the 2nd (Fig. 3, inset A). Stimulation of

the 1st or 3rd divisions similarly initiates this response in the 2nd division. Distant pick-up can be excluded as a factor in this observation.

Records obtained from the spinal V and main sensory nuclei are shown in Fig. 4. Recordings from these two regions differ. The delayed potential from spinal V has much the same configuration as that recorded peripherally but with a shorter delay following the initial spike (<1 msec.). The three main crests are again evident (Fig. 4, a, b, c).

The recording electrode for the main sensory nucleus (Fig. 4, MSN V) in this instance is at the lateral margin of the nuclear cell mass. The thresh-

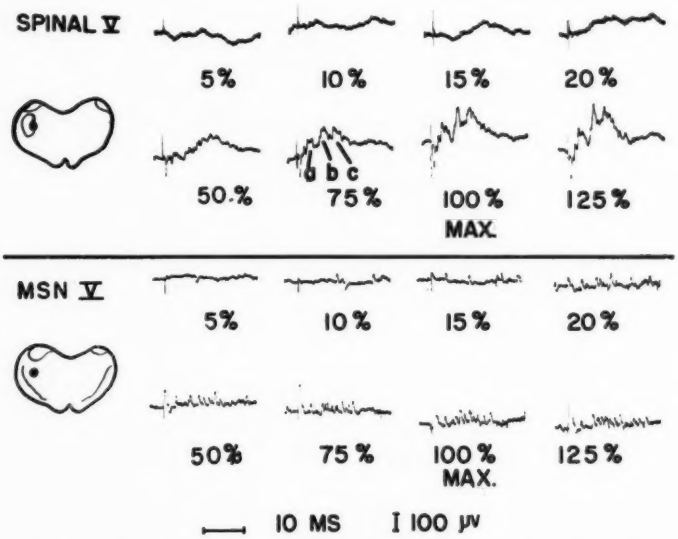

FIG. 4. Records contrast features of the response evoked in spinal V and in the region of the main sensory nucleus. A higher threshold and three crests in the delayed response in spinal V are evident.

old for an evoked response is distinctly lower here than in the spinal nucleus and the potential forms differ. When the main sensory nucleus electrode is accurately positioned, the first crest of the delayed response is large and has a low threshold, and maintains its initial form as stimulus strength is increased from threshold to maximum, suggesting its correlation with the large concentration of A fibers known to terminate in this nuclear mass (cf. Fig. 6, 1).

Fig. 5 further illustrates the form of these responses in spinal V. As the exploring electrode was inserted into the brain stem, the afferent spike (s) and the first crest (a) became clearly evident at a depth of 2 mm. At 2 5 to 3 mm., the response is clear and at 3.5 mm. almost absent. With decreasing voltages from maximum the changes in potential can be followed clearly

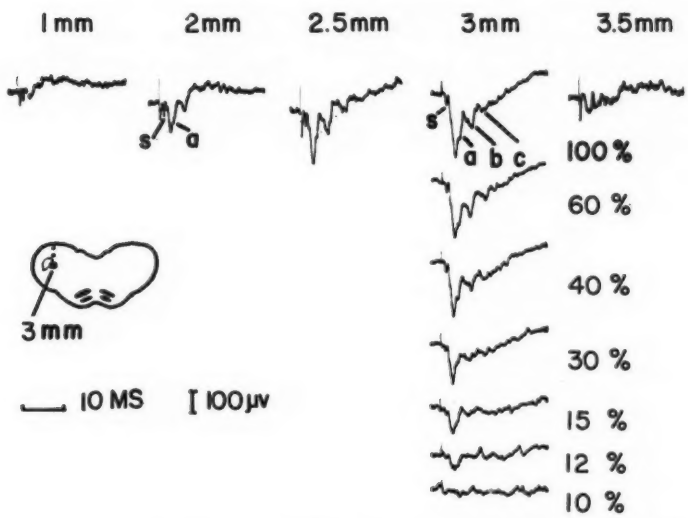

FIG. 5. The maximum response is indicated at depths in the medulla from 1 mm. to 3.5 mm. and at stimulus strengths from 10 per cent of maximum to maximum at a depth of 3.0 mm. The afferent spike (s) and characteristic three crests (a, b, c) of the delayed response in spinal V are clearly shown. The first crest (a) has a lower threshold than the succeeding crests.

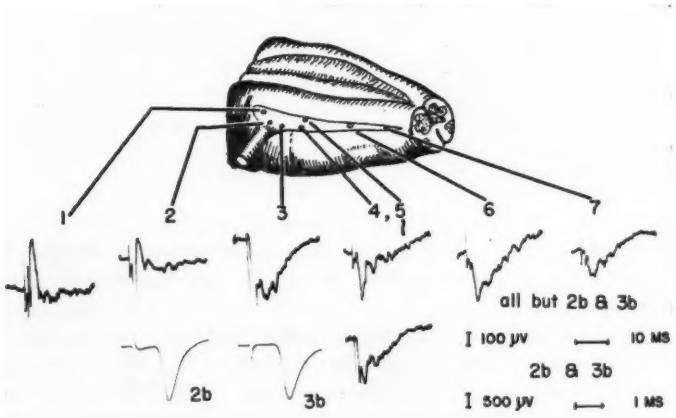

FIG. 6. The maximum response recorded from the main sensory nucleus (1), root entry zone (2, 3) and spinal V (4, 5, 6) varies significantly at each location. The high voltage afferent spike is pronounced in the root entry zone and lateral spinal V. The three crests of the delayed response are activated throughout the extent of spinal V. The response at the main sensory nucleus shows the afferent spike and only one crest in the delayed response.

at a depth of 3 mm. The 2nd (b) and 3rd (c) crests appear at higher thresholds than the first (a).

Fig. 6 demonstrates the maximum response to stimulation of the 2nd division as recorded in the spinal V and main sensory nuclei. The medial regions of the nucleus of the spinal V give little evidence of any afferent spike (5, 6, 7), while the two recordings from the root entry zone (2 and 3) and more lateral positions (4) show this feature clearly. The first crest is of high amplitude in the main sensory nucleus (1), where the 2nd and 3rd

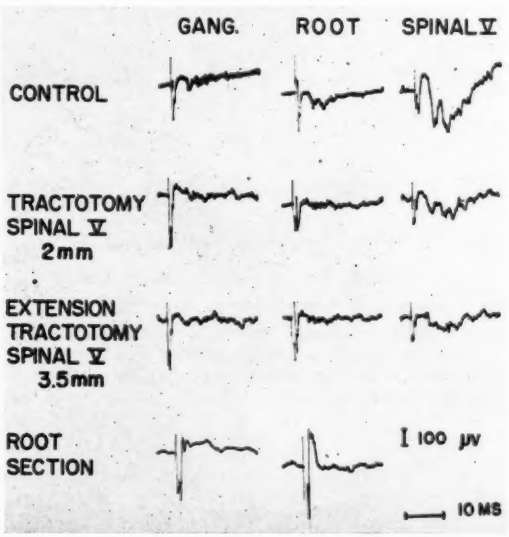

FIG. 7. Maximum response recorded in the Gasserian ganglion, posterior root and spinal V before tractotomy and following successively deeper incisions across spinal V 2 mm. above the obex of the fourth ventricle and caudal to the spinal V recording electrode. Similar records are shown from the Gasserian ganglion and posterior root following posterior root section between the recording electrode in the posterior root and brain stem.

crests are absent. In the spinal V, the first crest in this instance is still clearly activated as far caudal as the obex of the IV ventricle (7). In other preparations, this response has been recorded from the region of the tract of Lissauer at C_2.

This delayed response has been modified repeatedly in a constant manner and to a marked degree by a tractotomy of spinal V, 2 mm. above the obex and caudal to a recording electrode in the spinal V. The control records, as shown in Fig. 7, indicate maximum responses in the ganglion, root and spinal V. Following a shallow (2 mm.) tractotomy caudal to the spinal V recording electrode and 2 mm. above the obex, there is marked reduction in all elements of the delayed response. Although the delayed response persists, it is further reduced by extension of the incision to a depth of approxi-

mately 3.5 mm. Following root section the afferent evoked potential persists following stimulation, but the delayed response is completely abolished. These features indicate the central origin and *centrifugal* conduction of the delayed response.

That regional brain stem trauma alone is not responsible for these changes is demonstrated by control incisions (Fig. 8; 1, 2, 3). The delayed response was reduced only after incision of spinal V (Fig. 8; 4).

Much of this delayed response in the nerve and ganglion (but less of it in spinal V and but little from the ascending tract and main sensory nucleus)

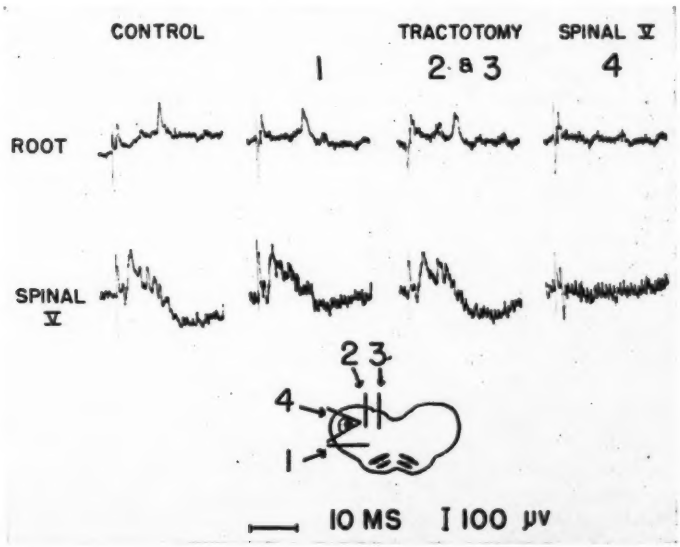

FIG. 8. Three control incisions into the medulla (1, 2, 3) caused no change in the trigeminal responses recorded in the posterior root and spinal V. A tractotomy of spinal V (4) 2 mm. above the obex of the fourth ventricle and caudal to the recording electrode in spinal V significantly reduced the response.

can be blocked by a tetanizing current as shown in Fig. 9. The fastest conducting large fibers contributing to the initial crest are maximally blocked by such repetitive stimulation. In the recovery phase the 2nd and 3rd components (b) of the delayed response do not reappear until 5 sec. after the initial crest (a) has regained maximum amplitude. The reappearance of the initial crest (conducted over large fibers), before the reappearance of the 2nd and 3rd crests (conducted over smaller fibers), indicates activation of the 2nd and 3rd crests by the larger fibers.

DISCUSSION

It is tempting to postulate on the basis of these findings, a mechanism centered in the spinal nucleus of the trigeminal, which under abnormal cir-

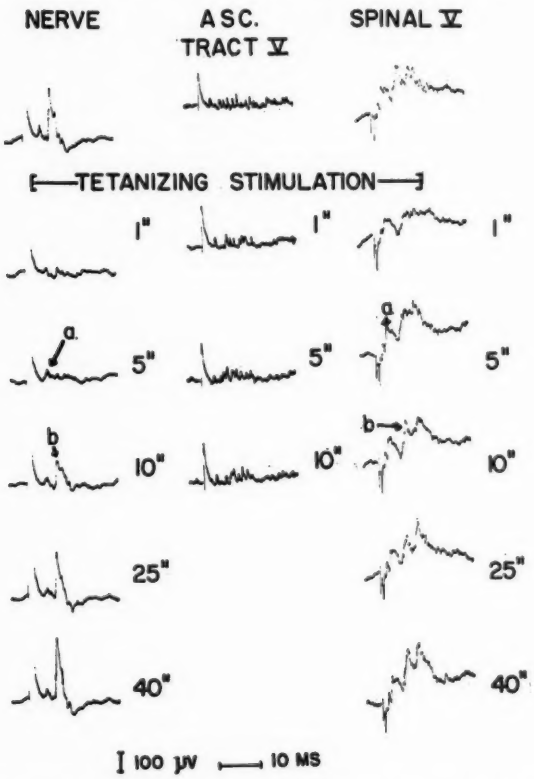

FIG. 9. Control records of maximum response in the 2nd division, ascending tract of V and spinal V are shown at the top of each column. Changes in the delayed response are shown at intervals from 1 to 40 sec., following a tetanizing current at 100 c.p.s. for 10 sec.

cumstances may be activated and play a role in the paroxysms of pain so characteristic of tic douloureux.

Several features of this response are compatible with such a supposition: (1) the delayed response is activated at low threshold by peripheral stimuli; (2) the delayed activity may be recorded in the 3rd division following stimulation of the 2nd division, suggesting a mechanism whereby stimulation in one peripheral trigeminal division might initiate pain referred to adjacent divisions of the trigeminal nerve; (3) the response is markedly reduced or abolished by tractotomy of spinal V, and is abolished by posterior root section; (4) this response is conducted over rapidly conducting pressure and touch fibers, which activate the initial crest of the delayed response. Some of these large touch axons are known to bifurcate on entering the brain stem. Their descending branches synapse in the spinal V nucleus where smaller fibers associated with pain conduction have numerous terminals.[8,13] (5) Blocking the rapidly conducting fibers with a tetanizing

current also effectively reduces the more delayed elements of the response until the first crest shows recovery. It would seem that the delayed response in the more slowly conducting pain fibers may be dependent to a degree upon afferent activation of the more rapidly conducting axons associated with touch for participation in this reflex activity.

In order to justify these suppositions which correlate the trigeminal dorsal root reflex activity with tic douloureux, additional requirements must be met.

1. Our observations on the trigeminal system show many features that are similar to the dorsal root reflex of spinal nerves reported by Gotch and Horsley,[3] Matthews,[5] Toennies[11,12] and Hursh.[4] This trigeminal reflex must differ significantly from the spinal dorsal root reflex, since tic-like syndromes of spinal nerves are not known. The following differences have been demonstrated in our records. Following a conditioning shock, the recovery phase of 150–300 msec. in this trigeminal reflex is shorter than the recovering phase of the spinal dorsal root reflex which may last more than one minute (4). The recorded amplitude of the trigeminal dorsal root reflex in our experiments was not greatly influenced by lowered body temperatures, whereas the amplitude of the spinal dorsal root reflex may show a five- to tenfold increase at temperatures of 30°C.[12] The activation of higher threshold axons increases the amplitude and modifies the form of this trigeminal reflex, whereas the addition of these elements to the spinal dorsal root reflex after the alpha response is maximum does not change significantly the form of the reflex potential.

2. It should be possible to produce in animals a syndrome similar to tic douloureux by changing this trigeminal reflex response to normal stimuli.

3. Before the proposed correlation between this trigeminal reflex activity and clinical tic douloureux can be accepted, it will be necessary to demonstrate changes in these reflex potentials in patients suffering from the disease in contrast to similar recordings from normal individuals. In addition some factor responsible for causing abnormal reflex responses must be demonstrated.

SUMMARY

1. Observations have been made on induced electrical activity in the trigeminal system of cats.

2. A correlation has been found between stimuli of large (touch) fibers and activation of small (pain) fibers in the caudal region of the spinal division of the trigeminal nerve.

3. The possibility is presented that a mechanism centered in the spinal trigeminal nucleus may participate under abnormal circumstances in paroxysmal trigeminal pain.

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CASE REPORTS AND TECHNICAL NOTE

OCHRONOSIS ASSOCIATED WITH DEGENERATION OF AN INTERVERTEBRAL DISC

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CASE REPORT

J. G., a 32-year-old white male, was admitted to the Hitchcock Hospital on Oct. 14, 1954 with the diagnosis of a protruded intervertebral lumbar disc.

Six weeks prior to admission, the patient lifted a loaded milk can on to the rear of a pickup truck and had immediate stabbing pain in the back. He had been in excellent health previously. He went to a chiropractor who relieved the pain in his back but caused severe pain in the right leg. There were no pains in the left leg and no disturbances of bladder. The pain in the right leg was made worse by coughing and sneezing. There were paresthesias in the right foot and toes.

A system review was negative save that he had been studied 8 years previously for aches of bones and joints and was thought to have had a "touch of polio." He was married and the father of three children. He had been in the U. S. Army for 5 years and had been in the Pacific Theater in World War II. He used no drugs. One older brother had had backaches for a long period of time but these had been attributed to "arthritis."

Examination. General physical findings were normal. His posture was normal but flexion of the spine in the lumbar region was limited to 10°. Walking on heels and toes was normal. There was severe spasm of the lumbar muscles. The right ankle jerk was absent. The other deep reflexes were normal. Motor and sensory findings were normal bilaterally. Babinski responses were plantar bilaterally. Straight leg raising on the right side was limited to 30° and there was a positive Lasègue sign on this side. The findings in the left leg were normal.

Red and white blood cell counts and differential white cell count were normal, and the Mazzini reaction (syphilis) was negative. The initial urinalysis showed a slightly positive reaction for sugar but no acetone or diacetic acid; two later urinalyses gave normal findings. No abnormality of color was noted. Roentgenograms of the chest and spine were negative except for narrowing of the lumbosacral space. A myelogram done elsewhere disclosed a defect at the lumbosacral space on the right side.

Operation. On Oct. 15, 1954 the patient underwent a laminotomy at the right lumbosacral space under spinal anesthesia. The laminae and the yellow ligament were normal. The posterior longitudinal ligament was markedly elevated and pressing on the 1st sacral root; the ligament was white in appearance. An incision into the ligament disclosed a black-appearing disc which was degenerated and soft (Fig. 1). This was easily removed in large pieces. A partial laminectomy at the 1st sacral segment was done and loose black cartilaginous material was found under the root, lying free in the canal. The root was well decompressed and closure was accomplished uneventfully.

Postoperative Course. There was transient inability to void. An indwelling catheter was utilized for 3 days. The urine was noted to be black upon standing, and reduced Benedict's solution. Ochronosis was established as the diagnosis. Closer examination of the patient failed to show any signs of the disease except for minor discoloration of the ears. Extensive roentgenographic investigation of the lumbosacral spine, hips, shoulders, hands and knees disclosed no abnormality.

He was free from pain after the 2nd postoperative day. He was placed on 100 mg. of ascorbic acid and citrus flavinoid compound three times daily although we did not anticipate any change in the color of the urine. He was given a course of strengthening exercises and was discharged on the 12th postoperative day. He remained symptom-free although he has recently passed a kidney stone and this problem is under investigation.

His older brother was investigated and found to have alcaptonuria and roentgenographic changes typical of ochronosis of the spine. The cartilages of the ears, nose and sternum were

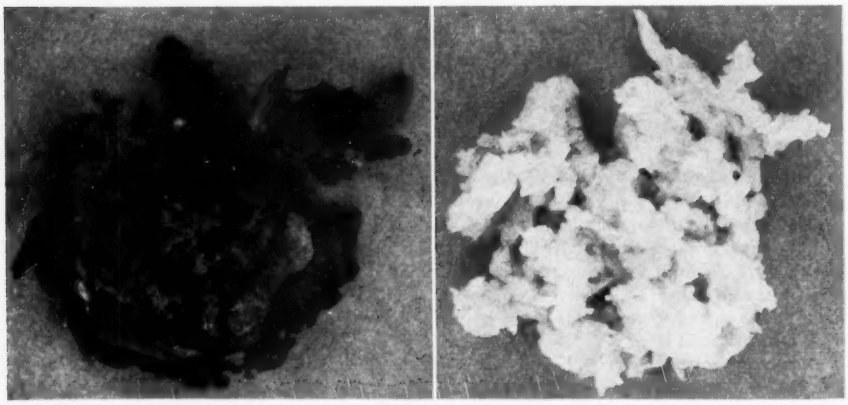

FIG. 1. Intervertebral disc removed in present case (*left*) contrasted with usual surgical specimen of a disc (*right*).

very black and his perspiration was dark-colored and stained his clothes. He was bothered by pain in the lumbodorsal region of the spine. The disease was not found in other siblings or children.

DISCUSSION

Ochronosis is a term used by Virchow[23] in 1866 to designate a condition in which the cartilages of the body appear black rather than white. The microscopic appearance of the granules in the cartilages was ochre rather than black. No mention was made in the article about the urine appearing black on exposure to the air; but Boedeker[4] in 1858 first recognized a substance in urine which could combine with oxygen in an alkaline medium to form a substance which he termed "Alcapton." The term alcaptonuria was derived from this. Wolkow and Baumann[24] in 1891 first recognized this compound as homogentisic acid. This compound appears in the urine when either of the two essential amino acids, phenylalanine and tyrosine, is incompletely metabolized. Homogentisic acid also appears in the urine of those having phenylpyruvic oligophrenia, tyrosinosis, or ascorbic acid deficiency.[5]

Homogentisic acid does not discolor acid urine but causes the urine to appear black or brown if alkaline.[1] It reduces Benedict's solution, does not ferment yeast, and does not rotate polarized light. This enables one to differentiate it from glucose and thus avoid confusion with diabetes mellitus. The occurrence of this compound is attributed to failure of the enzyme system to convert homogentisic acid into fumarylacetoacetic acid and eventually to fumaric acid and acetoacetic acid.[18]

Virchow[23] found the ochre pigment deposited in the eyes, skin, and hands, as well as the cartilages of the feet, ribs and large joints. It has been identified in many

tissues such as the heart, blood vessels, sweat glands, urinary, endocrine, and reticulo-endothelial systems.[14] Calcium is deposited with this pigment in the skeletal system.[11,19]

Changes occur in the spinal column beginning about the third decade. Lichtenstein and Kaplan[14] observed in 2 necropsied cases that the vertebral column was rigid and there were extensive changes in the region of the intervertebral disc. Exostotic changes were found with lateral protrusion of the disc and associated osteoarthritis. Parts of the disc had disappeared. Pigment was found in the annulus and nucleus pulposus and in the longitudinal ligaments. Bone was only slightly involved and the trabeculae showed osteoblasts and lacunae stuffed with pigment. However, Oppenheimer and Kline,[15] in one case, found flattening of the bodies of the vertebrae.

Degenerative changes were found in the coronary arteries by Osler;[16] and Young[25] reported that calculi of the prostate gland may be based on this condition. Smith[20] first described spots of pigment in the cornea near the temporal and nasal limbi as seen in the slit lamp. Sutro and Anderson[22] indicated that free bodies in the knee joint were caused by premature degeneration of the articular cartilage from the accumulation of homogentisic acid. Gross and Allard[10] first described the deforming arthritis of the larger joints in alcaptonuria.

Söderbergh[21] reviewed roentgenograms of 4 cases and found hypertrophic changes in vertebrae, hips and shoulder girdle. Pomeranz *et al.*[17] (the first to make this diagnosis solely from X-ray) found universal calcification of the discs which appeared as elliptical, opaque wafers in all the intervertebral joints, most strikingly in the lumbar area. Although these findings are considered characteristic of the disease, they were not present in our case.

Alcaptonuria is compatible with long life.[8] There may be no manifestation of the disease other than discoloration of the urine until the second or third decade. It occurs most commonly in males and is transmitted as a recessive trait.[9,12] Eventually, in most cases, extensive discoloration of the cartilages of the body will develop.[6] The perspiration and cerumen may be black.[2] There seems to be no specific effective therapy although many compounds such as ascorbic acid, cortisone,[3] and insulin have been tried in an attempt to convert homogentisic acid to its breakdown products. Klein and Bloch[13] were enthusiastic about parenteral liver therapy but this is not accepted by other authors.

It is clear that ochronosis can be responsible for destruction, calcification and pigmentation of intervertebral cartilage. Our case suggests that this process is one of the mechanisms that may lead to degeneration and rupture of an intervertebral disc. Although we are sure they must exist, we have not been able to find a report of a patient in whom a black intervertebral disc was removed at operation. Eisenberg[7] referred to a case of ochronosis which came to spinal fusion but the intervertebral discs were neither described nor biopsied.

In retrospect we have of course speculated as to why the preoperative urinalyses did not lead to the recognition of alcaptonuria. We suspect that the pertinent factors were the acidity of the urine and the length of interval between collection and examination. We believe that a preoperative laboratory diagnosis could usually be made if alcaptonuria is specifically suspected.

SUMMARY

1. The operative removal of a pigmented, protruded intervertebral disc in a patient with the metabolic evidences of ochronosis is reported.

2. The metabolic, roentgenologic, and stigmatic findings in ochronosis are briefly discussed.

3. Radicular symptoms related to ochronotic degeneration of an intervertebral disc may be present without the roentgenological findings or stigmata characteristic of this disease.

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INTRACRANIAL BOECK'S SARCOID TUMOR RESEMBLING MENINGIOMA*

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The numerous eponyms and terms that have resulted from the study of sarcoidosis over the years reflect the protean clinical manifestations of this disease entity. Historical reviews[14,22] reveal that in 1875 the first clinical report of a case was made by Hutchinson. In 1889 Besnier designated the skin changes "lupus pernio." Boeck (1899) described the histologic features of the cutaneous lesions and introduced the term "sarcoid." Heerfordt (1909) reported the uveoparotid fever syndrome, which later became identified as a manifestation of sarcoidosis. Schaumann (1914) was first to recognize the systemic nature of the disorder and suggested the term "benign lymphogranuloma." Osseous lesions, previously recognized by other observers, were established as part of the generalized disease by Jüngling in 1919.

Though extensive investigations as yet have failed to determine the etiology of sarcoidosis, clinical aspects have advanced impressively, and the cutaneous, visceral and lympho-hematopoietic manifestations have become well established.[2,5,11,15,16,20,21,23,29–32] Neurologic expressions of the disorder, not widely appreciated, are of particular interest. Involvement of the nervous system is relatively uncommon, and intracranial sarcoidosis simulating brain tumor is decidedly rare, only one case being cited by Essellier, *et al.*[7] in their extensive review of the literature in 1951. Additional cases of sarcoid tumor were recorded by Everts[8] in 1947 and by Höök[13] in 1954.

The present report is concerned with the successful surgical removal of a solitary, circumscribed intracranial tumor resembling a meningioma from the floor of the middle fossa. It was identified histologically as a sarcoid granuloma.

CASE REPORT

History. A negroid male, aged 19, on active duty in the U. S. Navy, was admitted to the hospital on July 18, 1954, because he had suddenly lost consciousness while aboard ship.

His illness began in July, 1953, when he was bothered by a roaring noise in his right ear. This was continuous and occasionally associated with headache and slight vertigo. In January, 1954, he was hospitalized for treatment of an infection of the right ear, and it was noted that he had polyps in the right external auditory canal. His symptoms continued and he returned to the hospital in April, 1954, when the polyps were surgically removed.

He then began to suffer from almost continuous right-sided headaches with intermittent nausea and vomiting. In addition he had frequent, brief episodes of slight mental confusion associated with unpleasant olfactory hallucinations. Distinct feelings of familiarity occurred at times, even when he was in a strange environment. In the month prior to admission he had two spells, characterized by automatic behavior with subsequent amnesia for his actions. He was found walking aimlessly about the ship, seemingly in a dazed state, and when attempts were made to restrain him he reacted in a maniacal, belligerent manner and finally lost consciousness.

* The opinions or assertions contained herein are the private ones of the authors and are not to be construed as official or reflecting the views of the Navy Department or the naval service at large.

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Examination. General physical findings were normal. Neurologic examination revealed bilateral papilledema of 4 D. There were small hemorrhages about both discs. Visual fields showed enlarged blind spots. The remainder of the cranial nerves were intact. The motor system showed good power throughout, although on prolonged repetitive movements the left extremities fatigued sooner than the right. There was mild increase in muscle tone in the left extremities. Coordination was normal. Sensation was unimpaired. The deep tendon reflexes were active and equal except that the left quadriceps response was slightly greater than the right. Plantar stimulation produced flexor responses of the toes bilaterally. The left superficial abdominal reflexes were consistently less active than the right.

Routine blood studies and urinalysis gave normal findings. Blood serology was negative. Roentgenogram of the chest showed no abnormalities. Skull roentgenograms revealed an

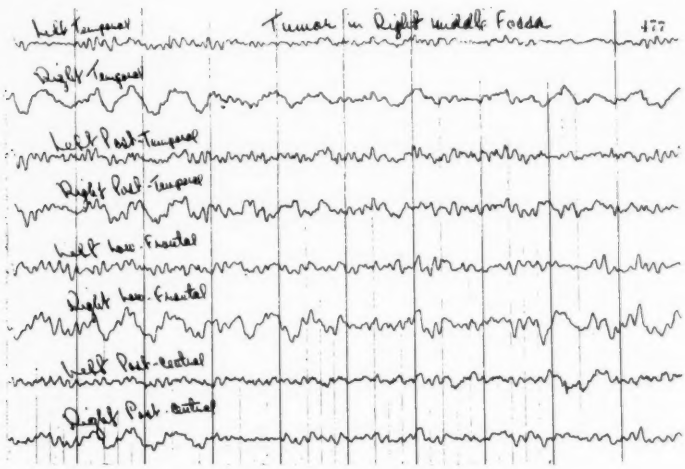

FIG. 1. Electroencephalogram showing high voltage slow activity in the right temporal and low-frontal areas.

enlarged sella turcica with demineralization of the clinoid processes. Lumbar spinal puncture showed a pressure of 280 mm. of clear CSF. The fluid contained no cells; sugar was 45 mg. per 100 cc., chloride 128 mEq. per liter, globulin 3+, and total protein was 100 mg. per 100 cc.

Electroencephalogram was disturbed by gross focal changes, consisting of higher potentials, slow activity of $1\frac{1}{2}$ to 4 per sec. frequency, irregularity of form and asynchrony which appeared in the anterior temporal, mid-temporal and low frontal areas on the right side (Fig. 1). Right percutaneous carotid arteriography revealed displacement of the middle cerebral artery upward and to the left, indicating the presence of a mass in the right temporal area (Fig. 2).

Operation. Craniotomy was performed on July 26, 1954. The tumor was not immediately evident on the surface of the temporal or posterior frontal lobes but was located by use of the Selverstone-Robinson probe counter, the patient having received intravenous radioactive phosphorus the previous day. Counting in the right temporal lobe delineated an area in which there was a ratio of 70 to 100:1 counts which extended from directly below the middle temporal gyrus medially for a distance of about 5 cm. Removal of a small portion of the inferior right temporal lobe revealed the tumor lying on the floor of the middle fossa. The mass was about the size of a goose egg, measuring $7\times5\times5$ cm., and extended medially almost to the mid-line. Removal of the tissue was accomplished by punch and electrocautery loop. The

tumor had a definite capsule, was not invading brain substance, and strongly resembled meningioma. It was fastened very firmly over an area of at least 1 square inch on the floor of the middle fossa. Most of the tissue attached to the floor of the fossa was removed.

Pathologic Examination. Gross. The submitted surgical specimens of the right middle fossa tumor consisted of moderately firm pieces of grey tissue. *Microscopic.* Sections of the tumor stained with hematoxylin-eosin showed dense granulomatous tissue composed of numerous small discrete and confluent nodular masses of epithelioid cells separated by lymphoid tissue (Fig. 3a). There were frequent multinucleated giant cells associated with the

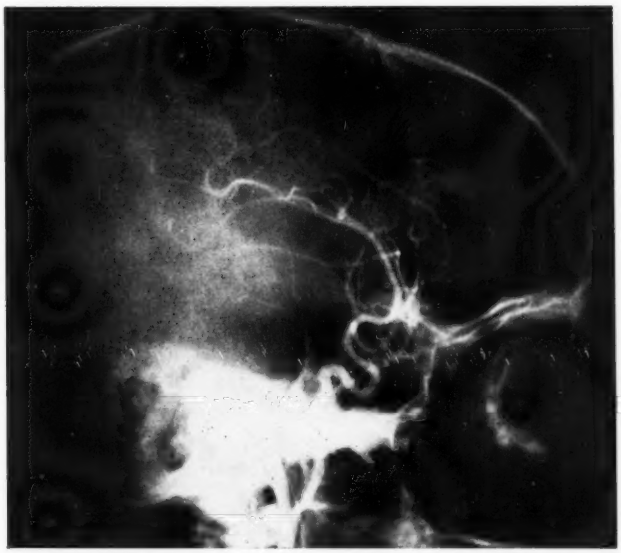

FIG. 2. Arteriogram demonstrating upward displacement of the right middle cerebral artery.

epithelioid cells. The giant cells had an abundant pink cytoplasm and many small round peripherally arranged nuclei (Fig. 3b). No specific etiologic agents were found. Acid fast stains were negative. There was no evidence of malignancy. Sections of the adjacent brain tissue showed some vacuolization and moderate gliosis as well as marked perivascular lymphocytic infiltration in a few places. The changes in the brain were presumably secondary to the adjacent granulomatous process. *Pathologic diagnosis:* Boeck's sarcoid granuloma.

Postoperative Course. Recovery was uneventful except for a mild hemiparesis of the left extremities for 1 month, after which no neurologic abnormalities could be detected. As a result of the histologic diagnosis a number of studies were performed in an attempt to detect any other manifestations of sarcoidosis. Intradermal tuberculin test (1:100) was negative. The Kveim skin test resulted in a negative response. Serial roentgenograms of the chest showed no abnormalities. Roentgenograms of both hands and feet were normal. Sedimentation rate was normal; plasma proteins, 8.2 gm. per cent; albumin, 4.0; globulin, 4.2; serum calcium, 9.0 mg. per 100 cc.; inorganic phosphorus, 4.4 mg. per 100 cc.; alkaline phosphatase, 2.8 Bodansky units. Sections of lymph nodes exised from the left supraclavicular area revealed no abnormalities.

Since discharge in November 1954, the patient has been doing full-time work at an airstation and has remained asymptomatic.

DISCUSSION

The diagnosis of sarcoidosis was not considered preoperatively in this case. At craniotomy the tumor gave the appearance of a meningioma. Subsequent investigation failed to reveal any evidence of systemic disease except for elevation of the serum globulins. Therefore, it cannot be determined conclusively whether the lesion represented an isolated sarcoid reaction or was a localized manifestation of generalized sarcoidosis. An increase in the globulin fraction of the serum proteins, like the negative reaction to intradermal tuberculin,[34] is commonly associated with

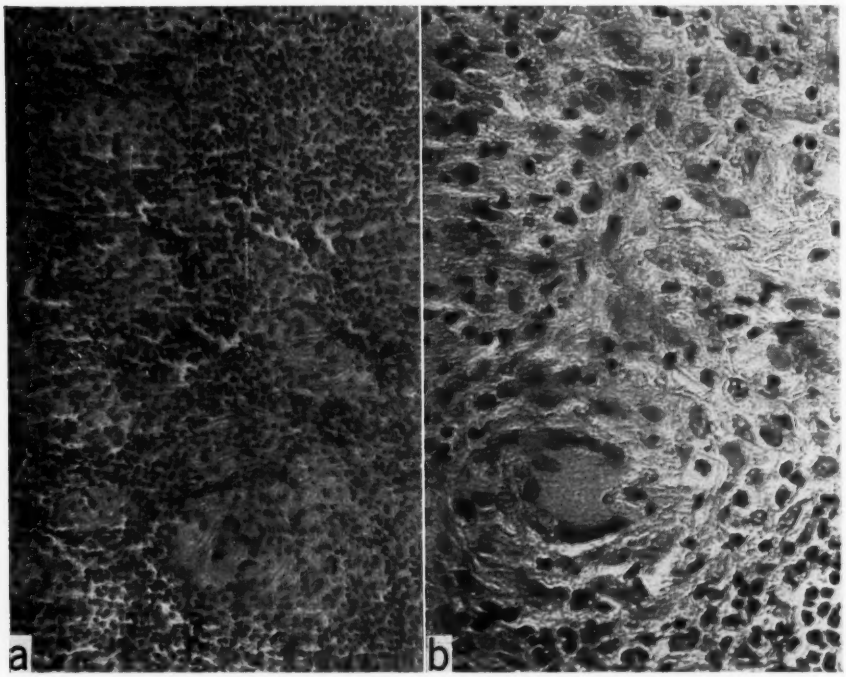

FIG. 3. Photomicrographs of tumor. (a) Dense granulomatous tissue composed of numerous smal nodular masses of epithelioid cells separated by lymphoid tissue. Hematoxylin-eosin stain, ×125. (b) Single Langhans giant cell; no evidence of necrosis, ×537.

Boeck's sarcoid but is not a pathognomonic finding. In regard to the Kveim skin test, various investigators have reported on the incidence of negative reactions in patients with proved sarcoidosis, with figures ranging from 8 to 35 per cent.[33]

The technic of employing radioactive phosphorus and a needle probe counter for brain tumor localization has been established as a distinct diagnostic aid[10] and in this case was of considerable value. The ratio between the count obtained in the area of the tumor and that obtained in the uninvolved brain varied from 70:1 to 100:1.

It is believed that the mass may have been enveloped by thinned dura mater

rather than by a true capsule, such as that which was described in the case report of Coleman and Meredith.[3]

Many recorded cases of sarcoidosis have lacked neurohistologic verification, the nature of the nervous system lesions being assumed on the basis of proved sarcoid elsewhere in the body.[17,18,28,36] Paresis of the facial muscles is the commonest sign of peripheral nerve implication, although any of the cranial nerves may be affected,[9] and there may be widespread neural involvement. The clinical expression of intracranial sarcoidosis is variable and depends upon the site of the infiltration. Diabetes insipidus has been a prominent symptom in about one-third of the recorded cases,[25] while convulsions, visual field defects, papilledema, spastic hemiplegia, hemianesthesia, cerebellar signs, mental aberrations, disturbances of sleep, headache, aphasia, and a variety of other cerebral signs and symptoms also have been described.[18,19,25,28,36]

Most of the pathologically established cases of central nervous system sarcoid have shown variable degrees of meningoencephalomyelitic infiltration, particularly of the basal structures. Cases of autopsy-proved meningoencephalitis caused by sarcoidosis have been reported by Erickson, Odom, and Stern,[6] Essellier, Koszewski, Lüthy, and Zollinger,[7] and Ricker and Clark.[27] Colover[4] recorded a case of posterior fossa arachnoiditis with fourth ventricle and cerebellar adhesions associated with sarcoid infiltration. Naumann[24] described sarcoid pachyleptomeningitis in an infant. Hypophysial infiltration characterized the case reported by Tillgren,[35] while the case of Gjersøe and Kjerulf-Jensen[12] showed involvement of the hypothalamus but not of the pituitary. Coleman and Meredith[3] recorded a case in which an encapsulated pituitary mass was surgically removed; it was initially reported as diffuse tuberculosis of the pituitary gland, but subsequent histologic study established the diagnosis of Boeck's sarcoid. Aszkanazy[1] recorded 5 cases: 1 with meningomyelitis of the upper thoracic cord; 1 with meningoencephalitis of the right pre- and postcentral gyri; 1 with pituitary invasion; and 2 with sarcoid tissue extending from the optic chiasm into the floor of the third ventricle. In 3 of these cases the disease could not be demonstrated elsewhere in the body. A case presented by Lenartowicz and Rothfeld[19] in 1930 and by Reis and Rothfeld[26] in 1931 revealed extensive sarcoid involvement of the cerebellum, infundibulum, floor of the third ventricle, both caudate nuclei, optic chiasm and nerves, anterior commissure, right olfactory bulb and gyrus rectus, left lenticular nucleus and left temporal lobe.

There have been very few reports of isolated tumors. Höök[13] described one patient with systemic sarcoidosis in whom a small tumor arising in the choroid plexus filled the upper portion of the inferior horn of the right lateral ventricle. It was extirpated and grossly resembled a choroidal papilloma; microscopic study established the diagnosis of sarcoid. Another case of generalized sarcoidosis presented by Höök lacked neurohistologic verification but there was clinical evidence of an expanding mass of the left frontal lobe and Sylvian fissure which disappeared under conservative medical management; such spontaneous and often dramatic remission of signs and symptoms is a characteristic feature of sarcoidosis. A patient of Everts[8] had a large left occipital lobe tumor which was successfully removed and histologically proved to be sarcoidosis; similar lesions were found in the posterior nasal pharynx, and roentgenograms of the chest revealed a left hilar mass. The case of Uehlinger (cited by Colover[4] and by Essellier *et al.*[7]) clinically demonstrated a mid-

brain syndrome; at autopsy there was a circumscribed mass at the base of the brain which histologically was sarcoid granuloma.

Emphasis has been placed on the cerebrospinal fluid changes in sarcoidosis.[13,25] Pleocytosis and an increase in protein content should arouse suspicion of sarcoid of the central nervous system in cases in which there is evidence of the disease elsewhere. A decrease in sugar content has also been reported. The spinal fluid in the present case contained no cells, elevated protein and lowered sugar content.

SUMMARY AND CONCLUSIONS

The case reported here clinically represented an expanding lesion of the right temporal lobe as manifested by olfactory hallucinations, psychomotor seizures, *déjà vu* phenomena, papilledema and slight left-sided signs of pyramidal tract dysfunction. Skull roentgenograms revealed enlargement of the sella turcica and demineralization of the clinoid processes. Electroencephalogram showed changes compatible with a gross irritative focus in the right temporal area. Elevation of the right middle cerebral artery, as demonstrated by carotid arteriograms, was indicative of a mass in the temporal lobe. At operation a large tumor with the gross appearance of a meningioma was found projecting upward from the floor of the middle fossa. It compressed the inferior portion of the temporal lobe, appeared to be encapsulated, and did not invade the substance of the brain. It was completely removed and histologically proved to be sarcoid granuloma. Subsequent investigation directed toward identification of systemic disease was negative except for elevation of the serum globulins.

Sarcoidosis of the central nervous system, though relatively uncommon, should be considered as a diagnostic possibility in patients with neurologic abnormality, including signs of cerebral tumor, who show evidence of sarcoid elsewhere in the body. Rarely, an intracranial lesion may represent the only discernible expression of the disease process.

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ARTERIOVENOUS MALFORMATION OF THE HEAD OF THE CAUDATE NUCLEUS

REPORT OF A CASE WITH TOTAL REMOVAL

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Vascular lesions protruding into the lumen of the ventricles, either from the roof or from the structures of the floor, are infrequently mentioned in the literature. Bassett[1] reported a patient (Case 5) who appeared to have had an intraventricular vascular anomaly arising from the corpus callosum and draining through the internal cerebral veins to the great vein of Galen. No air study was performed and ligation of the anterior cerebral artery distal to the anterior communicating artery was the method of treatment employed; no neurological sequelae were noted. Scott, Simril and Seaman[5] described a patient (Case 18) with an arteriovenous malformation in the left hemisphere extending into the left lateral ventricle, fed by the supracallosal artery, and draining into the vein of Galen. The patient died of "cerebral edema and thrombosis" after surgical extirpation.

The following case is of interest in that an arteriovenous malformation arising from the head of the caudate nucleus and protruding into the left lateral ventricle was successfully removed by a transventricular approach.

CASE REPORT

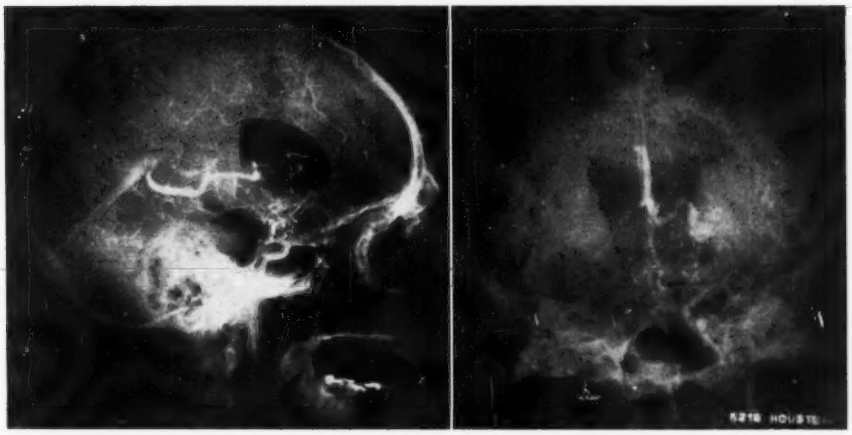
V.A. #37031. B.G.S., a 38-year-old white male, was admitted to the Veterans Administration Hospital, Houston, Texas on Feb. 8, 1954, 2 weeks after the onset of apparently severe subarachnoid hemorrhage. Past history was noncontributory, except for intermittent, poorly localized headaches of 8 years' duration. In 1951, 3½ years before admission, he had what was then called an "epileptic convulsion" in which he lapsed into unconsciousness for a period of 2 hours, without apparent residua; further details are unavailable concerning this episode. Three weeks before admission the headaches became severe and were associated with numbness and tingling of all extremities. Two weeks before admission he suddenly announced that he was "about to have a stroke" and immediately thereafter became comatose. He was admitted to another hospital, where it was noted that the cerebrospinal fluid was grossly bloody and his B.P. was 200/100. He remained unconscious for approximately 1 week, requiring oxygen and parenteral fluids; congestive heart failure was treated by digitalization. There was marked fluctuation in his blood pressure, with numerous episodes of severe hypertension. Upon regaining consciousness, he was irrational, had difficulty in swallowing and had a minimal right hemiparesis. Vomiting unaccompanied by nausea was frequent. He was transferred to the Veterans Administration Hospital.

Examination. His pulse rate was 82 and B.P. was 100/70. He was fairly alert, being oriented for person, but not for time and place. Both Kernig and Brudzinski signs were positive. There was minimal right-sided weakness. Sensory findings were normal, as were the cranial nerves; there was no papilledema. No bruit was audible.

Laboratory Data. Hemogram was normal. Bleeding time was 2 minutes, clotting time 4½ minutes, and platelet count 352,000. Blood chemistries, consisting of sodium, potassium, chloride, carbon dioxide, urea nitrogen, and fasting sugar all were within normal limits.

Kline was normal. CSF pressure on Feb. 12, 1954 was 140 mm. The fluid was xanthochromic and contained 6 WBC and 4 RBC per c.mm.; protein was 144 mg. per cent; colloidal gold curve was normal; Wassermann test was negative. Roentgenograms of skull and chest revealed no abnormalities. ECG was normal. EEG done on Feb. 9, 1954 showed grossly abnormal and paroxysmal generalized frontal dominant slow bursts with some left-sided lateralization.

Course. The patient was thought to have had a subarachnoid hemorrhage. Cerebral angiography was performed under local anesthesia on Feb. 16, 1954, injecting each common carotid artery and taking multiple serial exposures. Injection of the left side revealed a small cluster of vessels deep in the left hemisphere, presumably in the left lateral ventricle, which drained almost immediately into the left internal cerebral vein, great vein of Galen,



FIGS. 1 and 2. Combined pneumoencephalogram and angiogram with left common carotid artery injected. (*Left*) Left lateral brow-up view. Mass of vessels in left lateral ventricle and head of caudate nucleus draining into left internal cerebral vein and, in turn, into great vein of Galen and sinus rectus. Note that lesion is anterolateral to choroid plexus, with which there is no apparent connection. (*Right*) Frontal projection (slightly angulated). Feeding vessels from M1 portion of middle cerebral artery are clearly seen, as well as large draining vessel towards midline into left internal cerebral vein.

and straight sinus. Injection of the right side, without compression on the left, resulted in filling of the arteriovenous malformation. During the procedure the patient suddenly became confused and his blood pressure precipitously rose to 200/100.

On Feb. 23, 1954, under surital anesthesia, a 40 cc. pneumoencephalogram was performed, this being followed by another left carotid angiogram with anteroposterior and lateral single views. These (Figs. 1 and 2) showed the relationship of the vascular lesion to the ventricular system; it was thought that the lesion arose in the head of the caudate nucleus and protruded into the left lateral ventricle.

Operation. On Mar. 9, 1954, a left frontal craniotomy was performed (Fig. 3). A plug of cortical tissue was removed anterior to the coronal suture and the left lateral ventricle was entered. The arteriovenous malformation was seen in the head of the caudate nucleus 2 cm. from the anterior pole of the ventricle, protruding into the ventricular lumen. This portion of the lesion consisted of numerous loops of vessels, which seemed to drain into the left internal cerebral vein via a curved vessel posteromedial to the body of the lesion. No feeding vessels were visible, specifically, none from the choroid plexus. The ventricular system was deformed; there was complete communication between the atria of both ventricular cavities, with the

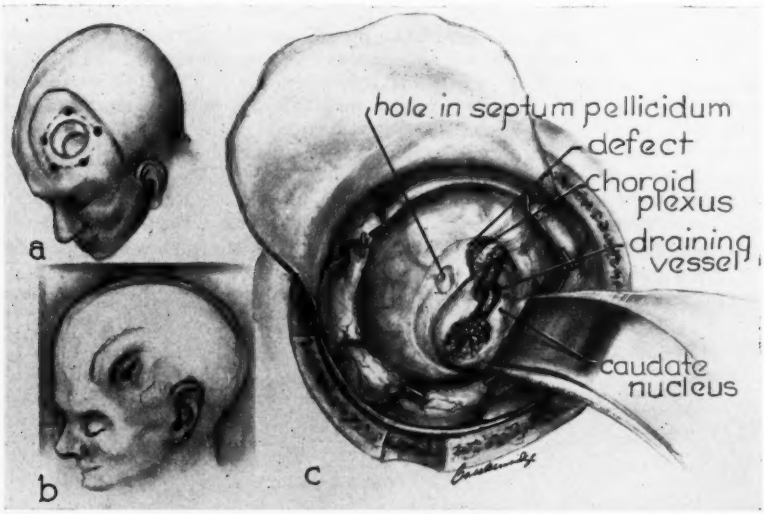


FIG. 3. Artist's conception of lesion and operative approach. The ventricular defect refers to the intercommunication of both ventricles in the atrial region. Note the visibility of the choroid plexus in the tela choroidea of the third ventricle and its course into the lateral ventricle, all of which were visualized at operation.

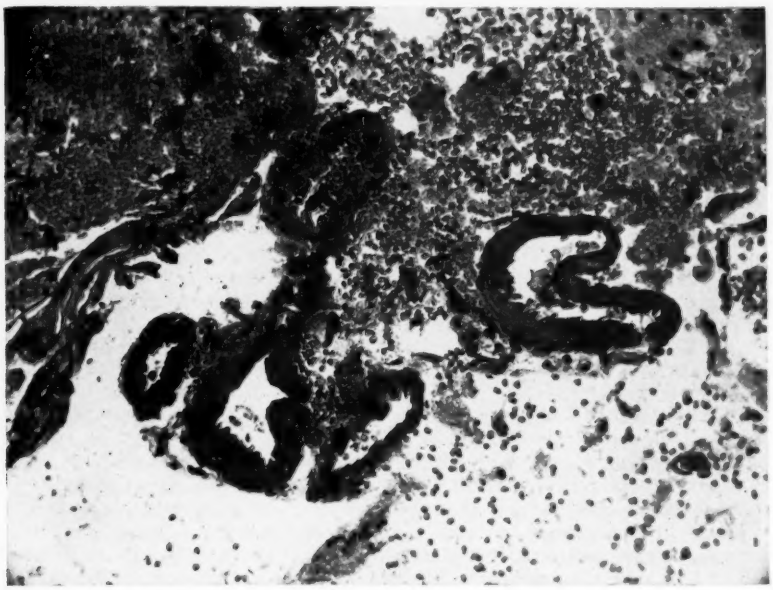
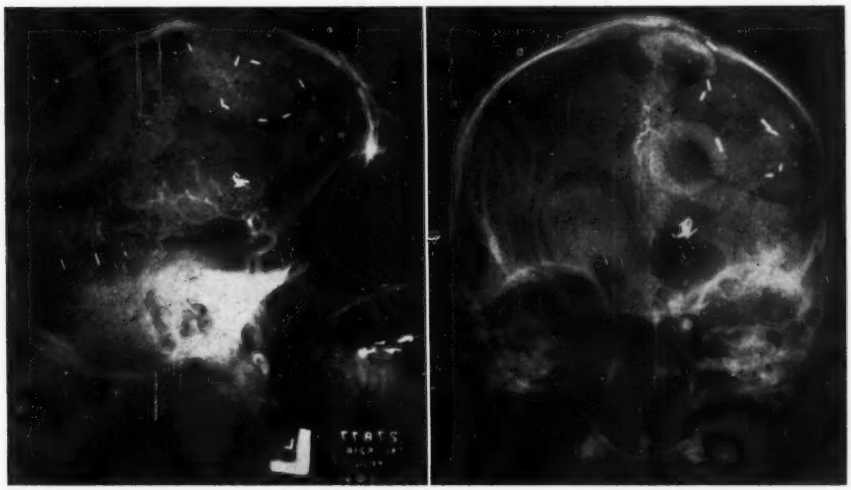


FIG. 4. Photomicrograph of operative specimen, showing communicating racemose blood spaces with endothelial lining, a delicate media, and absent adventitia, ×40.

choroid plexus and venous structures in the roof of the third ventricle clearly visible; the complete course of the choroid plexus through the foramina of Monro into the lateral ventricles could thus be seen. There was also a perforation in the remaining septum pellucidum anteriorly. The ependymal lining of the ventricular system showed a greenish-yellow discoloration, probably caused by old hemorrhage.

Under induced hypotension (Arfonad, or RO 2-2222), the lesion was initially attacked by placing silver clips on the visible vessels, with the hope of obstructing the main feeder. This did not prove feasible and because of persistent bleeding, the dissection was carried into the head of the caudate nucleus for approximately 1 cm., clipping the vertically coursing feeding vessels arising from the middle cerebral artery. The lesion was completely excised, clipping the draining vessel last, this being collapsed and devoid of blood at this time.

Two interesting episodes occurred during anesthesia, one prior to institution of hypotension. This consisted of a sudden drop in pulse to 48 with a rise of systolic blood pressure



FIGS. 5 and 6. Postoperative left common carotid angiograms. (*Left*) Left lateral view, showing absence of lesion. (*Right*) A-P view. Note small lesion just proximal to clips, which has not been removed, and which may represent another small vascular anomaly.

to 170 mm. Hg, associated with ECG evidence of ventricular extrasystoles and partial heart block; the response to intravenous atropine sulfate was immediate, with reversion to normal rhythm. At the close of the procedure, when the blood pressure was normal, there was a drop of the pulse to 28, with bigeminy and trigeminy, and a marked hypertension of 220/170. The blood pressure gradually lowered and the pulse became regular in response to intravenous pronestyl.

Pathological Report. The surgical specimen (S-434-54) revealed large thin-walled blood spaces communicating with one another, compatible with the diagnosis of hemangioma (Fig. 4).

Postoperative Course. This was uneventful, except for a minimal right hemiparesis, more marked in the leg, which rapidly and completely cleared. The patient was ambulatory on the 10th day. His mental confusion, poor memory, tendency to perseverate, and emotional lability, all gradually cleared.

Left-sided angiography was performed on April 7, 1954 (Figs. 5 and 6), revealing the absence of the previously shown arteriovenous malformation. A small dilated vessel (Fig. 6),

just proximal to the clips, may represent a residual portion of the lesion, or a separate small anomaly.

Fifteen months after operation the patient showed no abnormal neurological findings. There was minimal reduction in performance on psychological testing.

COMMENT

The use of combined pneumoencephalography and angiography served to delineate the relationship of the arteriovenous malformation to the ventricular system. It should be noted, however, that less of the lesion was floating free in the left lateral ventricle than would seem apparent from the radiographs. It was unfortunate that a complete study of the ventricular system was not performed (this was thought too risky at the time, only brow-up anteroposterior and lateral views being taken), since it would undoubtedly have revealed the intercommunication of both lateral ventricles, as previously described.

In discussing the theories of origin of the cavum septi pellucidi and the cavum vergae, Wolf and Bamford[6] support Corning's[2] view that the cavum vergae is probably a portion of the interhemispheric fissure whose lateral boundaries are the medial aspects of the cerebral vesicles ventral to the caudally extending corpus callosum. The defect present in this case, although showing no evidence of either of the above anomalies, may have been caused by a failure of development of the medial portion of the cerebral vesicles, and consequent intercommunication of the portions of the ventricles. The defect would therefore be on a congenital basis, rather than caused by the secondary results of massive intraventricular bleeding.

The marked fluctuations in cardiovascular responses, prior to and during surgery, likewise remain unexplained. Both the severe hypertension and transient heart block of varying degree may well have been related to the site of the pathological lesion, since autonomic changes have been elicited by stimulation in neighboring areas (the "septo-hypothalamic" region of Heath, *et al.*[3,4]).

SUMMARY

The successful removal, without significant clinical residua, of an arteriovenous malformation in the head of the left caudate nucleus and left lateral ventricle is reported.

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OSSIFYING CHRONIC SUBDURAL HEMATOMA

REPORT OF A CASE

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Ossifying chronic subdural hematoma is a rare condition. In the literature only 4 such cases are known to have been studied histologically, and in only 1 of these was true bone found.

REPORT OF CASE

A 30-year-old man was admitted to the Neurosurgical Clinic of the Hospital "Evangelismos" in July 1953 stating that in 1941 he had been struck on the head by a member of the occupying forces and, after a short period of unconsciousness, he was well and did not complain until a year before admission when he began to suffer from headaches. A month later he had the first general epileptic seizure with foaming at the mouth and unconsciousness. Following this seizure he was never well and complained of dizziness and debility. Seizures continued to occur about twice a month. The patient entered the neurological clinic of the Hospital (Dr. Patricios) from which he was transferred to our department.

Examination. General and neurological findings were normal.

Roentgenograms of the skull showed a plate of calcification beneath the inner table on the right, extending from the frontal to the parietal region (Fig. 1). The sella turcica was normal in size and shape and there was no erosion of the clinoids. An encephalogram failed to fill the ventricles.

The cerebrospinal fluid was under normal pressure; cells, protein and colloidal gold reaction were normal.

Angiography showed an interruption of the outline of the vessels at the site of the calcification. The diagnosis was uncertain but we decided upon an operation.

Operation. On Sept. 13, 1953, under endotracheal (trilenium) narcosis, an osteoplastic craniotomy was made in the right frontoparietal region. The dura mater appeared normal but there was palpable beneath it a hard plate which gave the impression of bone. This hard subdural mass was movable although slightly adherent to the dura mater, which was carefully dissected back and reflected toward the longitudinal sinus. The calcified mass extended up to the sinus and it was perforated by veins which entered the sinus. The exposure was sufficient to display all the calcified mass which was adhering to the arachnoid. After ligation of the veins the mass was freed from the thickened arachnoid and removed.

Course. The patient made an uneventful recovery.

Pathological Report. The specimen consisted of a hard material resembling bone and it was slightly concave (Fig. 2a). It measured 10×6 cm., and 4 mm. in thickness. The inner side was mottled and very irregular and some of these portions were white. On microscopic examination the specimen was found to be composed of ripe osteoid tissue with formation of Haversian canals surrounded by osteoblasts. A roentgenogram of the specimen is shown in Fig. 2b.

DISCUSSION

Our first thought was that there must have been calcification of a chronic hematoma, though one could not exclude the possibility of a heterotopic osteoma. But the study of the case, and the publication of a similar case by Chusid and de Gutiérrez-Mahoney[1] strengthened our opinion that it was an old hematoma that had become ossified. As has been accepted by the majority of neurosurgeons, after a head injury there is sometimes formed a small hematoma from rupture of veins

FIG. 1. Preoperative roentgenogram showing plate of calcification extending from frontal to parietal region.

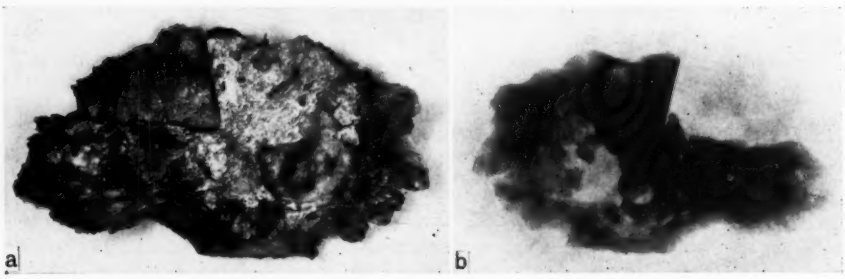

FIG. 2. (a) Subdural mass removed at operation. (b) Roentgenogram of same.

and, after a period of time which may vary from a few weeks to years during which there are no symptoms, the hematoma may grow larger by the absorption of cerebrospinal fluid and produce symptoms. Calcification of the chronic subdural hematoma is frequent but not its ossification. Chusid and de Gutiérrez-Mahoney, who reviewed the literature, found 3 cases of histologically verified ossification within a chronic subdural hematoma but only 1, described by Wertheimer and Dechaume, contained typical true bone. The present report describes a second case in which on microscopic examination true bone was found in the mass removed by operation.

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CONGENITAL SPINAL EXTRADURAL CYST

CASE REPORT AND REVIEW OF THE LITERATURE

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One of the most favorable spinal lesions from the standpoint of both surgical extirpation and recovery of neurological function is the extradural cyst of non-traumatic origin. It is also one of the rarest of intraspinal lesions. To date, including the present case, 34 proved cases have been recorded in the world literature to our knowledge. These are listed chronologically in Table 1.

The first verified report was that of Collins and Marks[5] in 1915, which described a patient operated upon by Dr. Alfred S. Taylor. The second case, reported by Mixter[21] in 1932, was somewhat atypical in that the cyst was multilocular. The article by Elsberg, Dyke and Brewer[9] in 1934, reporting 4 cases, described the clinical syndrome and suggested the congenital origin of these lesions. These authors were apparently unaware that their Case 4 had been previously reported by Collins and Marks[5] (Case 2). In 1937, Cloward and Bucy[3] recorded a case and carefully reviewed the history, findings and roentgenograms in 9 previously reported cases. In 8 of the 10 cases, the cyst was located in the thoracic region, and each of the patients had more or less marked radiological stigmata of kyphosis dorsalis juvenilis (Scheuermann's disease). Cloward and Bucy advanced the hypothesis that the extradural cysts, by interfering with the venous drainage of the vertebral bodies, caused the kyphosis dorsalis juvenilis. The case reported by Meredith[20] is the only one in which the cyst occurred in the cervical region. Some question arises (and was raised by the author) as to whether this cyst was caused by trauma.* In 1947, Swanson and Fincher[27] reported 2 cases of extradural cysts. The lesion in the first patient, a 20-month-old child, was an extensive arachnoidal cyst; it protruded through a dural defect extending from the 2nd lumbar to the 1st sacral segment and contained several roots of the cauda equina. It is probable, as the authors concluded, that this lesion was traumatic in origin. In the case of their second patient, however, it seems more likely that the lesion was a congenital extradural cyst, the symptoms of which were precipitated by "trauma" (sneezing). The latter case is included in this review.

CASE REPORT

A 29-year-old radio repairman of Chinese extraction was admitted to the San Francisco Veterans Administration Hospital for investigation of backache of 6 years' duration. On subsequent close questioning, he recalled that when he was 12 or 13 years of age, he had had transient episodes of paresthesias, numbness and weakness of the right lower extremity after lengthy automobile trips. These symptoms disappeared after several years.

* Cohen's[4] patient had an extradural cyst at T8, probably asymptomatic, and an extradural "neuro-(sympatico)blastoma" at T10; the latter probably caused the spinal cord compression.

Six years prior to admission, the patient noted the gradual onset of episodes of dull, nonradiating low backache after sitting for any prolonged period. The pain was not severe; it was aggravated by straining at stool but not by bending or coughing. One year later, occasional dysesthesias were noted in the lateral surface of the right thigh and calf and in the toes of the right foot. About 3½ years before hospitalization, slowly progressive weakness of the right lower extremity developed, mainly distally, and progressive wasting of the right calf became apparent during the following year. About this time, mild weakness and dysesthesias of the left lower extremity appeared. There was slight difficulty initiating and terminating micturition when the backache was present, but no actual urinary incontinence or retention.

While in the U. S. Army from 1942 to 1946, the patient served in Africa, China and India, but did not contract any illness.

Examination. The patient was a thin, short Chinese male. No spinal deformities were noted, and there was a full range of motion of the back without pain, tenderness or muscle spasm. Rectal sphincter tone was good. On the right side, the quadriceps reflex was slightly hyperactive and the Achilles reflex was absent. Left quadriceps and Achilles reflexes were normal. Abdominal reflexes were active and equal; plantar reflexes were flexor. There was hypalgesia of the lateral portion of the right foot extending to the lateral malleolus. Position sense and vibration perception were intact. There was marked atrophy of the intrinsic musculature of the right foot, with a pes cavus deformity, and the circumference of the right calf was 1½ inches (3.75 cm.) less than the left. Thigh measurements were equal. There was moderate weakness of the right extensor hallucis longus, extensor digitorum longus and gastrocnemius-soleus group, and slight weakness of the right peronei and tibialis anticus.

Laboratory Data. Complete blood count, urinalysis and blood serological tests for syphilis were within normal limits. Free acid was present in the gastric contents. Roentgenograms of the lumbosacral spine were interpreted as normal. Pantopaque myelography was carried out and the myelograms were interpreted as revealing a slight defect on the right at the 4th lumbar interspace. It was reported to us subsequently, however, that the dye was not run up to the upper lumbar region. There was no evidence of spinal subarachnoid block. The cerebrospinal fluid was clear and colorless, with no cells, a protein of 67 mg. per cent and a negative Wassermann.

1st Operation. On May 27, 1954, the right 3rd, 4th and 5th lumbar interspaces were explored. The operative report stated that no evidence of disc protrusion or nerve root compression was found.

Course. Within 2 weeks there was a moderate increase in strength of the right lower extremity and the patient was discharged.

He returned 3 weeks later, complaining of increasing weakness of the *left* lower extremity. The right quadriceps reflex was slightly hyperactive and the Achilles reflex was absent as before. The left quadriceps reflex was normal, but unsustained left ankle clonus was now present. The left plantar reflex was flexor, but there was no response on the right. The "hamstring" group, tibialis anticus, extensor hallucis longus, and peronei, were slightly weak bilaterally, but worse on the right side. Hypalgesia had extended to involve the lateral surface of the right leg. Vibratory perception was absent in the toes bilaterally, and was slightly decreased in the feet. Position sense was intact.

Pantopaque myelography was repeated. Cerebrospinal fluid dynamics were normal, although the fluid appeared faintly xanthochromic and the protein content was 136 mg. per cent. Ascent of the dye was completely blocked at the level of the 2nd lumbar vertebra.

2nd Operation. On July 20, 1954 a thoracolumbar laminectomy was performed, and the laminae and spines of the 11th and 12th thoracic and 1st lumbar vertebrae were removed. As the laminae were removed, no epidural fat was encountered, and the ligamenta flava were adherent to what appeared to be dura mater. In removing the ligaments the supposed dura mater was inadvertently opened and clear fluid ran out. When the laminectomy was completed, it became apparent that an extradural cyst was present dorsal to the true dural sac, and this was actually what had been opened. The upper pole of the cyst narrowed to a small cul-de-sac which was loosely adherent to the dura mater at the level of the 11th thoracic

vertebra. The lower pole of the cyst was at the 2nd lumbar vertebra as determined by measurement with a catheter. When the cyst was fully opened and all fluid drained from it, compression of the jugular veins caused bulging of the dura mater. No fluid entered the cyst, however, indicating the absence of a patent communication between the subarachnoid space and the cyst. The cyst could be readily peeled from the dura mater, but laterally was densely adherent to the epidural veins in several places. Consequently, the dorsal and ventral walls of the cyst were excised, leaving behind fragments of the lateral extensions where adherent. In removing the ventral surface of the cyst, a small opening in the dura mater appeared at the origin of the dural sleeve for the right 12th thoracic nerve root. This was presumably the obliterated pedicle from which the cyst arose.

Microscopic Examination. The cyst wall was composed mainly of fibrous tissue, without evidence of inflammatory reaction or hemorrhage, suggestive of dura mater and arachnoid.

Postoperative Course. This was uneventful. Motor power gradually improved and there was complete relief of the backache.

When the patient was examined on Oct. 20, 1954, motor power was intact except for moderate weakness of the right extensor hallucis longus and slight weakness of the right tibialis anticus. The left Achilles reflex was active, the right was absent. Both quadriceps reflexes were absent, but reflexes were active and equal in the upper extremities. Plantar reflexes were flexor. There was slight hypalgesia of the dorsum of the right foot. Position sense was intact. The right calf measured 30.6 cm. in circumference, the left 32.8 cm.

DISCUSSION

The report of Elsberg *et al.*[9] established the typical syndrome of the spinal extradural cyst, and Cloward and Bucy[3] pointed out the relationship of these lesions and kyphosis dorsalis juvenilis. Very little has been added to the clinical picture since these papers appeared. It is generally agreed that these lesions have a congenital origin (other types of cysts, e.g., parasitic and traumatic, can be excluded by microscopic examination). Elsberg *et al.*[9] proposed two possible theories of origin, namely, (1) a congenital diverticulum of the dura mater, or (2) herniation of the arachnoid through a congenital defect in the dura mater. Hyndman and Gerber[14] postulated that the cysts arise from cell rests. This latter theory seems unlikely, since in at least 11 of the reported cases there was a direct communication between the cyst and the subarachnoid space. In the others, it is probable that a communication existed originally but was obliterated. We believe this to have been the situation in our patient, for although no direct communication was demonstrated, a small opening appeared in the dura mater when the cyst wall was dissected free.

A review of the 33 cases (Table 1) in which details are available reveals the following facts: The average age of the patients was 22.6 years, with 1 patient aged 6 years, 18 in the 10–19 year group, 6 in the 20–29, 2 in the 30–39, 5 in the 40–49 year group, and 1 patient aged 51. Twenty-three were male and 10 female. The cyst was located in the thoracic region in 24 instances, thoracolumbar in 6, lumbar in 2, and cervical in 1. Thirteen patients gave a history of spontaneous remission or improvement with conservative therapy (e.g., immobilization, hyperextension). The average duration of symptoms was 35 months. Ten patients had symptoms for less than 6 months, 7 for 6 months to 1 year, 10 for 1 to 5 years, and 6 for over 5 years. The cyst communicated with the subarachnoid space in 11 cases; no such communication was observed in 10 cases; and communication was undetermined in 12. A partial or complete block of the spinal subarachnoid space, as determined by the Queckenstedt test, was present in 18 cases. In 6, no block existed, and the results were not stated in 9 reports. Slight to marked evidence of kyphosis dorsalis

TABLE 1

*Summary of 34 cases of congenital spinal extradural cyst**

Authors	Age	Sex	Duration of Symptoms	Location of Cyst	Remission or Improvement: Conservative Therapy	Communication with Subarachnoid Space	Subarachnoid Block (Queckenstedt)	Kyphosis Dorsalis Juvenilis	Comments
1. Collins & Marks[5] 1915	15	F	2 yrs.	T3-6	Yes	?	?	?	Subsequently reported by Elsberg *et al.*[9] (Case 4).
2. Mixter[21] 1932	26	M	13 yrs.	T3-7	Yes	?	No	Yes	Multilocular
3. Elsberg *et al.*[9] 1934	12	M	3½ yrs.	T5-12	Yes	No	Yes	Yes	Initial diagnosis of multiple sclerosis.
4. Elsberg *et al.*[9] 1934	15	M	3 mos.	T6-9	No	?	Yes	Yes	
5. Elsberg *et al.*[9] 1934	15	M	9 mos.	T4-9	No	?	Yes	Yes	
6. Lehman[18] 1935	12	M	3½ yrs.	T6-8	Yes	Probable	Yes	Yes	Initially improved with hyperextension.
7. Lehman[18] 1935	17	M	9 mos.	T6-10	No	?	Yes	Yes	
8. Peet & Kahn[22] 1936 (Case 3)	12	F	2 mos.	T9-L4	No	?	Yes	Yes (Thoracic only)	Marked vasomotor changes, all extremities. Disapp. postop.
9. Cloward[2] 1937	43	M	2 yrs.	T10-L5	No	No	No	No	
10. Teachenor[28] 1937	—	—	—	—	—	—	—	—	No details available.
11. Kelly[16] 1937	15	M	5 mos.	T5-8	No	Yes	Partial	Yes	
12. Cloward & Bucy[3] 1937	20	M	4 yrs. Kyphos. 6 yrs.	T7-9	Yes	No	No	Yes	Temp. improvement with hyperextension +body cast.
13. Robertson & Graham[23] 1939	14	M	6 mos.	T8-11	No	?	Yes	Yes	
14. Turnbull[29] 1939	14	F	3 mos.	T6-11	Yes	Yes	Partial	Yes	Temp. improvement with extension on Bradford frame.
15. Meredith[20] 1940	41	M	8 mos.	C7	No	Yes	Yes	No	Only cervical cyst questionably caused by trauma.
16. Adelstein[1] 1941	15	F	5 mos.	T5-7	No	?	Yes	Yes	
17. Gros[11] 1942	14	M	1 mo.	T4-9	No	?	?	?	
18. Hartmann[13] 1942	14	M	1½ yrs.	T6-9	No	Yes	?	?	
19. Mayfield & Grantham[19] 1942	16	M	3 yrs.	T6-9	Yes	Yes	Yes	Yes	
20. Mayfield & Grantham[19] 1942	26	M	12 yrs.	T5-8	Yes	No	?	Yes	Initial presumptive diag.: multiple sclerosis.
21. Good *et al.*[10] 1944	42	M	8 yrs.	T8-L1	No	Yes	Partial	Yes	
22. Cohen[4] 1945	48	F	3 mos.	T8	No	?	Yes	No	Associated with "neuro(sympatico)blastoma."
23. Shenkin *et al.*[26] 1945	51	F	3 mos.	T7-9	?	No	?	No	Contained "oily fluid."
24. Shenkin *et al.*[26] 1945	13	F	2 yrs.	T6-8	?	No	?	No	
25. Hyndman & Gerber[14] 1946	13	M	8 mos.	T4-10	No	No	Yes	Yes mild scolios.	
26. Swanson & Fincher[27] 1947 (Case 5)	43	F	"several years"	T11-L2	Yes	?	?	No	
27. Turner[30] 1947	11	M	5 mos.	T6-11	Yes	Yes	Yes	Yes	Improved following spinal puncture.
28. Decker & Livingston[7] 1949	31	F	6 yrs.	L3	Yes	Yes	No	No	
29. Davis[8] 1949	18	M	9 mos.	T6-8	No	Yes	Partial	No?	
30. Hamlin *et al.*[12] 1949	25	M	2 yrs.	T12-L2	Yes	?	No	No	
31. Schreiber & Nielsen[25] 1950	27	M	2½ yrs.	L2	No	Yes	?	No	
32. Zoltan & Farago[31] 1951	6	F	1 yr.	T4-6	No	No	?	Yes	Overlying skin nevus.
33. Jacobs *et al.*[15] 1954	32	M	9 yrs.	T6-8	No	Yes	Partial	Yes mild	Cyst filled on myelography.
34. Wise & Foster 1955	20	M	16 yrs.	T11-L2	Yes	No	No	No	

* The patients reported by Krauss[17] and by Schlesinger[24] have been excluded from this review since their lesions were probably not congenital spinal extradural cysts. This has been pointed out previously by Cloward and Bucy[3] and by Mayfield and Grantham.[19]

juvenilis was present in 19 patients, absent in 11, and not stated in 3. In all instances of kyphosis dorsalis juvenilis, the cyst was located partially or completely in the thoracic portion of the spinal canal.

The problem of diagnosis of a spinal extradural cyst is complicated by certain peculiarities of the clinical picture. Pain is generally slight or absent, spontaneous remissions may occur, and the patient is often young. A diagnosis of multiple sclerosis was made initially in several cases (Elsberg *et al.*,[9] Mayfield and Grantham[19]). When the bony changes of kyphosis dorsalis juvenilis have been marked, a

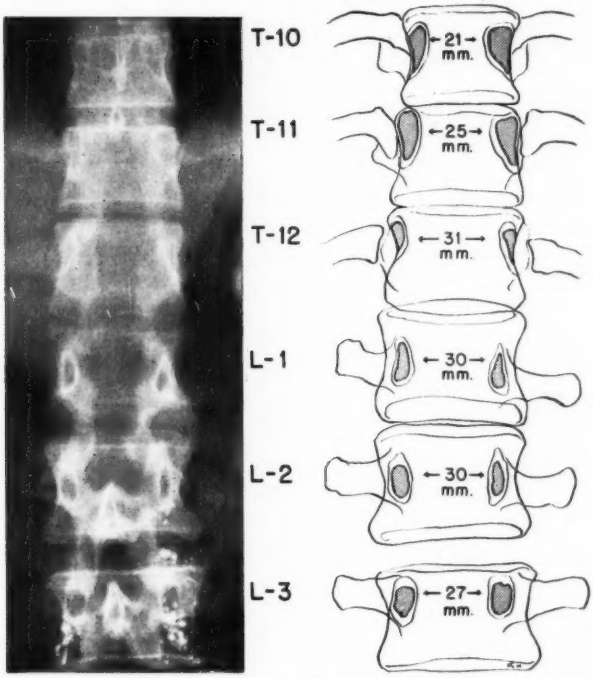

FIG. 1. (*Left*) Anteroposterior roentgenogram of thoracolumbar spine (postoperative). (*Right*) Tracing of same, with interpedicular measurements superimposed, and pedicles accentuated. Note enlarged interpedicular space of T12 and erosion of both T12 pedicles and left L1 and L2 pedicles.

diagnosis of tuberculosis of the spine has been made on several occasions, apparently confirmed at first by the improvement in paraplegia following hyperextension and immobilization of the patient's spine (Lehman,[18] Cloward and Bucy,[3] and Turnbull[29]).

When the cyst is located in the lumbar or thoracolumbar region, pain in the low back and lower extremities may occur. When weakness, sensory loss, and reflex changes are present in the lower extremities, the diagnosis of protrusion of a lumbar intervertebral disc may be suggested, as in our patient. This case illustrates once again the importance of *adequate* myelography, i.e., fluoroscopic visualization of the entire lumbar and lower thoracic portions of the spinal canal, in patients with the "lumbar disc syndrome."

In the case reported, marked atrophy of the musculature of the right foot and lower leg had occurred, with similar changes beginning on the left. Had the condition progressed much further, it might have simulated peroneal muscular atrophy (Charcot-Marie-Tooth). In any "atypical" case of peroneal muscular atrophy adequate myelography should be performed before this diagnosis is accepted.

Finally, careful examination of the spinal roentgenograms and measurement of the interpedicular spaces may establish the diagnosis. Elsberg *et al.*[9] first pointed out the widening of the interpedicular space and erosion of the pedicles caused by extradural cysts. Postoperative roentgenograms of the lower thoracic and upper lumbar spine in our patient indicated that both these abnormalities were present (Fig. 1). The interpedicular measurement of T12 was 31 mm. (extreme upper limit of normal is 30 mm.[8]) and there was definite erosion of the pedicles of T12. The 1st and 2nd left lumbar pedicles appeared slightly thinned as well.

SUMMARY AND CONCLUSIONS

1. A case of spinal extradural cyst, presumably congenital in origin and located in the thoracolumbar region, is presented.

2. Thirty-three patients with such lesions have been recorded previously in the world literature. The characteristic aspects of these cases are reviewed, and their differential diagnosis is discussed.

ADDENDUM

Since this paper was prepared, an additional case was described by Dr. Henry M. Cuneo at the combined meeting of the Southern California Neurosurgical Society and the San Francisco Neurological Society at Pebble Beach, California, February 26, 1955.

The patient was a 48-year-old male. Motor and reflex changes were more prominent than sensory loss. The pedicles of the 6th to 8th thoracic vertebrae were eroded. Scoliosis was present, centering at the 2nd lumbar vertebra. The cyst was located at the level of the 5th to 8th thoracic vertebrae, communicated with the subarachnoid space, and filled with dye during myelography. Postoperative improvement was reported to be slight.*

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A NEW METHOD FOR TRACHEOTOMY*

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A patent airway is of vital importance in every patient. The indications for tracheotomy in the treatment of respiratory obstructions have long been recognized. The technic is well established, and has been changed little if any during the past fifty years.

Tracheotomy as standardized is a surgical procedure, consisting of skin incision, dissection of the soft tissues, exposure of the trachea, incision of the cartilaginous rings and insertion of a silver tube into the trachea.

The standard operative method is satisfactory for elective tracheotomy for relief of a chronic tracheobronchial obstruction. As an emergency procedure it has several obvious disadvantages. Hospital facilities are required, as well as a complete set of surgical instruments. Considerable time is required, especially by those not familiar with the technic, which adds additional trauma to the patient already in critical condition.

Our interest in tracheotomy stems from the care of patients with serious acute head injuries. Associated fractures of the jaw or facial bones, as well as lacerations of the soft tissues of the face, mouth or pharynx result in excessive bleeding, much of which gravitates into the tracheobronchial tree, causing acute respiratory embarrassment. Spinal fluid escaping from basal skull fractures frequently adds to the obstruction.

A patient with a severe head injury is often deeply comatose, has lost his protective gag reflex and literally drowns in his own secretions. Lesser accumulations of blood and secretions in the bronchial tree may cause partial obstruction with secondary cerebral anoxia and increase in carbon dioxide sufficient to produce severe cerebral edema and a marked increase in intracranial pressure.

This vicious cycle can be interrupted only if the air passages are aspirated of all foreign material. The administration of nasal oxygen is of no value if the trachea or bronchi are obstructed. Aspiration through the nose is difficult as only occasionally can a soft rubber catheter be passed between the vocal cords into the trachea.

Tracheotomy in such patients is a life-saving procedure. The tracheotomy tube allows frequent adequate aspiration, assures a continued supply of oxygen, and prevents accumulation of carbon dioxide.

Many patients in dire need of tracheotomy are denied the benefits of an artificial airway because of the complexities of the present methods. If a simple, rapid means of tracheotomy were available, it no doubt would become a routine procedure, thus saving many hundreds of lives each year.

The method described below fulfills these requirements. The tracheotomy tube

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can be inserted rapidly and safely. No surgical instruments or hospital facilities are necessary. It can be used by any physician, or in the Armed Services could easily be used by hospital corpsmen.

METHOD

The instrument at the present time consists of four parts (Fig. 1):

1. A needle with a retractable barb that is first inserted to transfix the trachea (A). Thus the trachea can be elevated (B) against the overlying skin and lateral motion of the trachea can be prevented while the tube is being inserted.

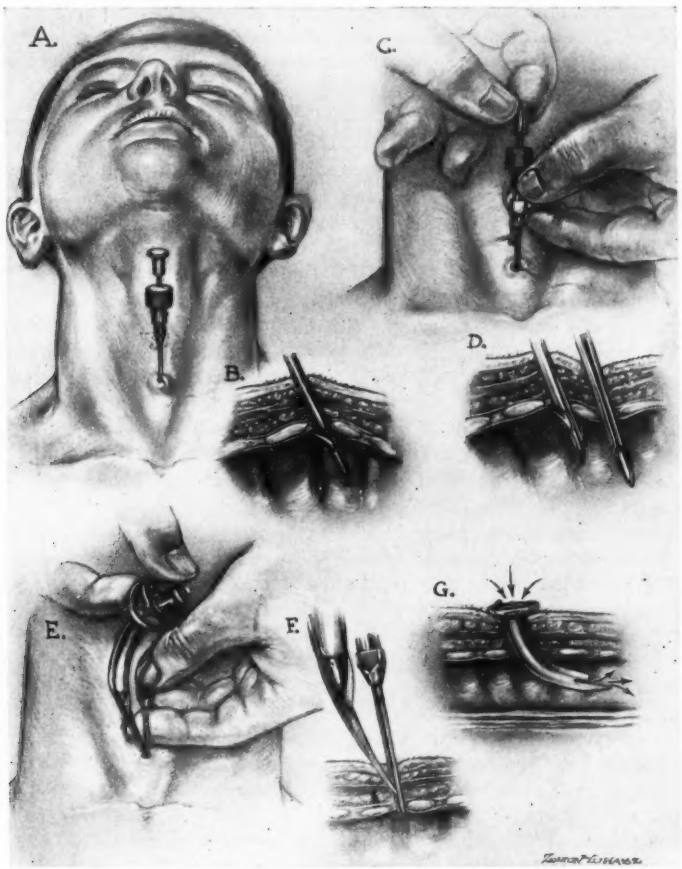

FIG. 1. See text for description.

2. A silver tracheotomy tube with three narrow slots in the tapered distal end.

3. A cutting trocar that slips through the tracheotomy tube and projects beyond the distal tapered end. The cutting blades are two in number. One is a long tapered spring steel blade sharpened on both edges. On the end of this blade is a small ball tip which prevents the tapered horizontal blade from piercing the posterior wall of the trachea. The second vertical

blade is short with a convex cutting edge placed at right angles to the long tapered blade.

4. A short #13 needle is required that has a portion of the wall ground away so as to leave a slot along the distal portion. The proximal end of this slot is enlarged to a circular opening large enough to admit the ball-like tip of the long tapered horizontal cutting blade.

Before introducing the tracheotomy tube, the patient is placed in a supine position on the table with the head hyperextended. The trachea is located and the first needle is passed through the skin into the lumen of the trachea (A). Pressure on the handle end of the trocar in this needle unlocks the barb flange which is opened by then pulling back on the end of the trocar. This flange protrudes as a blunt fishhook against the inside of the trachea. Traction on the needle now elevates and transfixes the trachea (B).

While traction is thus exerted, the second slotted needle is introduced through the skin into the lumen of the trachea (C). This needle is introduced about 2 cm. caudad from the first needle (D). The ball-like tip of the horizontal cutting blade is inserted through the spherical opening in this needle and the ball is passed down the lumen of the needle into the trachea (E). The blade itself slides in through the slot extending in the needle from the spherical opening to the tip (F). Once the long tapered horizontal cutter is within the trachea the needle is removed.

The cutting blades with the attached tracheotomy tube are then passed into the trachea. Once the tube is within the lumen of the trachea the cutters are withdrawn through the lumen of the tracheotomy tube leaving the patent tube in place to serve as an airway (G).

DISCUSSION

The instrument had been tested many times, using skin and subcutaneous tissues in the neck and elsewhere to determine the optimum length, shape and cutting surfaces of the horizontal blade. The short convex blade was tested with sections of trachea to judge the ease with which the right-angle incision could be made in the tracheal cartilage.

The most important step in the use of the instrument is the insertion of the #13 needle into the lumen of the trachea.

We were concerned with the accuracy of placing the tip of the needle, although it seemed almost impossible for one to miss the trachea considering the large diameter of the lumen as compared to the average subcutaneous vein or even the common carotid artery deep in the neck.

Once the tip of the needle is in the lumen it is impossible to introduce the tube and cutters anywhere except into the trachea since they both follow the slot extending the length of the needle. The ball tip on the horizontal cutter prevents the instrument, once it has entered the trachea, from escaping from the lumen.

Finally it was tried clinically and found to function much easier through living tissue. The problem of the needle solved itself. The needle proved to be large enough for a marked exchange of air through the spherical opening on the side of the needle. One could easily hear the exchange of air and thus be certain that the needle was properly located in the trachea. The cutters and tube entered the trachea with very little effort.

The first patient on whom a tracheotomy was done by this method had a severe acute head injury, complicated by marked anoxia caused by tracheobronchial obstruction from mucopurulent secretions. The patient was deeply comatose and seemed to have little if any cough reflex present.

The total time required for the introduction of a standard #6 tracheotomy tube was less than 30 seconds. The patient made a slow but satisfactory recovery. There was some redness and slight edema about the tube for a few days but not enough to be of any clinical significance. The tracheotomy was allowed to function a little over

2 weeks and then the tube was removed. The opening closed promptly and with far less scarring than usually follows the usual open method.

We have not employed this method in children because the models developed have incorporated the large #6 tracheotomy tube used for adults. Smaller sizes can be made for children and should function equally well. The fact that the trachea in children is less rigid than in adults should offer no problem since the cutting edges follow the course of the needle, the tip of which is already within the lumen of the trachea. Therefore, the action on the trachea is one of cutting rather than pressure.

We no longer use the needle with the retractable barb (Fig. 1 B) to transfix and elevate the trachea. We have found that the cutting blades and tracheotomy tube are readily passed into the trachea when the head and neck are in maximum hyperextension. The pressure required to introduce the instrument is exerted in a direction closely parallel to the long axis of the trachea, hence there is no tendency for the trachea to move laterally or to be compressed.

This instrument was developed for use in situations demanding immediate tracheotomy. From a neurosurgical standpoint, we have been concerned mainly with patients who have received severe head injuries.

We have employed the instrument in patients whose necks varied greatly in length and width. Even in patients with short, thick necks, there has been no difficulty in performing the tracheotomy.

The first tracheotomy by this method was done in December 1953. Since that time we have used it routinely without incident of bleeding, infection, tracheal injury or any other complications.

We are certain that technical improvements can and will be made on this basic model, but from a practical and clinical standpoint it already has afforded us a safe, simple and speedy method of insuring an adequate airway.*

* This instrument is in the process of being manufactured by the Sierra Madre Manufacturing Company of Sierra Madre, California. It is being licensed by the Research Corporation of America and will be in production within the next month or two.

BOOK REVIEWS

A TEXTBOOK OF NEUROLOGY. By H. Houston Merritt. *Philadelphia: Lea & Febiger*, 1955, 746 pp., 181 figs., and 128 tables. Price, $12.50.

This new textbook of neurology reflects the extensive experience of its author in clinical neurology and neuropathology. The work is extremely well organized and practical. Of great significance is the emphasis throughout the text of the relationships between neurology and internal medicine. The attempt has been made to present the neurological disorders according to their etiologic aspects. The chapters therefore have been divided into the subjects of infections, vascular lesions, tumors, trauma, developmental defects, degenerative diseases, diseases caused by toxins, metabolic disease, demyelinating diseases, and diseases and functional disturbances of unknown etiology, including epilepsy. Each particular entity is discussed with regard to etiology, pathology, incidence, symptoms, diagnosis, course and treatment.

There are numerous tables, well chosen illustrations and in general the style is succinct. The discussions of treatment are thorough, critical, and highly practical. Following each review of a disease entity there is an extensive up-to-date list of references. The usual sections on applied neuro-anatomy and physiology and examination of the nervous system are not present in this book, and the author states in his preface that, in a text such as this, the coverage of these subjects is usually inadequate and this material is better covered in special texts. However the pertinent aspects of neuro-anatomy and physiology and the disordered features seen on neurological examination are discussed when necessary throughout the text. One can agree with this method of approach in a modern textbook of neurology since it is more important to have a thorough and detailed coverage of the disease entities and problems of differential diagnosis and treatment. The various groups of disease have received proper emphasis since over half of the book consists of the sections on infections, vascular lesions, tumors, trauma and developmental defects. It might have been helpful, however, to have a section on aphasia, apraxia, and agnosia. Worthy of special mention is the extremely thorough and useful index. The value of this textbook improves on usage and it is highly recommended.

G. H. GLASER, M.D.

HISTOLOGICAL TECHNIQUE FOR INTRACRANIAL TUMOURS. By Dorothy S. Russell.

Professor Dorothy S. Russell's valuable book on histological technique in neurosurgery, originally published by the Oxford University Press in 1939, has long been out of print. It has been set up in mimeograph form and is now available for distribution. While it has been impossible to reproduce the illustrations, this is compensated for by revisions made by Professor Russell. The book is of the greatest help, not only because it gives directions for management of neurosurgical material, but also because it has a good deal of wisdom on the diagnosis of such material. It is warmly recommended to those preparing for neurosurgical Board examinations.

Copies can be obtained by sending a check for $2.00, made out to Larue D. Carter Memorial Hospital, to Miss Edna M. Fuller, Larue D. Carter Memorial Hospital, 1315 West Tenth Street, Indianapolis, Indiana, U.S.A.

ORVILLE T. BAILEY, M.D.

NOTICES

AMERICAN BOARD OF NEUROLOGICAL SURGERY

There has been much discussion by the Board regarding problems relating to residency training programs, and after giving the matter thought and consideration the Board has decided that the training program in neurological surgery should be changed so that four years of residency training in neurological surgery will be required beyond the one year of required training in general surgery. A rotating internship fulfills only six months of the requirement in general surgery, and the basic time beyond graduation is being increased to five years. After at least one year of acceptable training in general surgery, the individual may begin his training in neurological surgery, and this training must, as in the past, be progressive, with more and more responsibility being assumed by the individual as his training progresses. During these four years of training at least thirty months must be spent in actual contact with clinical neurological surgery. Upon completion of the training program the individual should have had adequate training in the basic sciences so that he will be able to pass the Board examinations in organic neurology, general surgery, neuro-anatomy and neurophysiology, neuropathology, neuroradiology, and neuro-ophthalmology. This change is being made because in the past the three-year training program has been so set up that during only two years of the training period was the trainee actually in contact with clinical neurological surgery, and the Board feels that this amount of time is inadequate. The additional year may be spent in either basic research, general surgery, or medical neurology, and the method of spending the additional year should be determined by the trainee and the chief of service during his training. This will allow considerable latitude in the training program which would be adjusted according to the individual's needs and the need of the service involved.

This change in the training program does not affect individuals who have already begun their training in neurological surgery, and since the change cannot be published officially until the Residency Number of the Journal of the American Medical Association comes out in the fall, it may be construed as applying to those individuals who begin their training as of July 1, 1956.

The American Board of Neurological Surgery held its thirty-second official meeting in St. Louis, Missouri, April 27–30, 1955. Examinations were held at the Washington University Medical School, and the following candidates were certified:

ALIFF, ROBERT C. 329 East Davis Blvd., Davis Islands, Tampa 6, Fla.

AMERONGEN, FREDERICK K. 4418 Vineland Ave., North Hollywood, Calif.

BICKERS, DONALD S. 710 Peachtree St., N.E., Atlanta, Ga.

BOEHM, WALTER E. Interstate Bldg., Chattanooga, Tenn.

CARTON, CHARLES A. Baylor University College of Medicine, Texas Medical Center, Houston, Texas

CHRISTOFERSON, LEE A. 435 Delendrecie Bldg., Fargo, N. D.

COOPER, JACK R. 665 New Brotherhood Bldg., Kansas City, Kan.

CROSBY, R. M. N. 11 East Chase St., Baltimore, Md.

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EVANS, WILLIAM G. 1910 Knoxville, Lubbock, Texas

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HEIFETZ, MILTON O. 6317 Wilshire Blvd., Los Angeles, Calif.

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ROSS-DUGGAN, JOHN K., II 1933 Pacific Ave., Long Beach, Calif.

SHENKMAN, SAMUEL 67 East 78th St., New York, N. Y.

SWAIN, GARRETT M. 1904 R St., N.W., Washington, D. C.

TOZER, RICHARD C. 3617 West 6th St., Topeka, Kan.

YUHL, ERIC T. 1486 Glendon Ave., Los Angeles, Calif.

ACADEMY-INTERNATIONAL OF MEDICINE

A completely revised fourth edition of *Professional Films* is being compiled. It will contain biographical data on authors of motion-picture films and information on the audio-visual activities of medical and dental schools and postgraduate teaching centers.

Authors of films are requested to write to the Academy-International of Medicine, 601 Louisiana Street, Lawrence, Kansas for questionnaires, or provide the film title and full name and address of any film author.

AMERICAN NEUROLOGICAL ASSOCIATION

At the 80th annual meeting of the American Neurological Association held in Chicago from June 13–15, 1955, the following officers were elected: President, Dr. J. M. Nielsen; President-Elect, Dr. H. Houston Merritt; 1st Vice-President, Dr. James W. Kernohan; 2nd Vice-President, Dr. Robert B. Aird; Secretary-Treasurer, Dr. Charles Rupp; Assistant Secretary, Dr. William F. Caveness.

CONGRESS OF NEUROLOGICAL SURGEONS

The next meeting of the Congress of Neurological Surgeons will be held at the Los Angeles Statler Hotel in Los Angeles, California, Oct. 27–29, 1955. The Secretary is Dr. Philip D. Gordy, 1007 Delaware Avenue, Wilmington, Delaware.

HARVEY CUSHING SOCIETY

The 23rd annual meeting of the Harvey Cushing Society was held in Quebec, Canada, May 16–18, 1955. The following officers were elected: President, Frederic Schreiber, Detroit; President-Elect, Leo M. Davidoff, New York; Vice President, James G. Lyerly, Jacksonville; Secretary-Treasurer, David L. Reeves, Santa Barbara; Historian, Louise Eisenhardt, New Haven; member of Executive Committee, Harry Wilkins, Oklahoma City. Mrs. Robert H. Pudenz, Pasadena, is President of the Ladies Auxiliary, and Mrs. John E. Webster, Detroit, is President-Elect.

JOURNAL OF NEUROSURGERY

The *Journal of Neurosurgery* is devoted to the publication of work relating primarily to neurosurgery, including studies in clinical neurophysiology, organic neurology, ophthalmology, radiology and pathology. Short articles on unusual cases and on technique, special instruments or equipment that might be useful to others in the field are also acceptable.

MANUSCRIPTS should be addressed to Dr. Louise Eisenhardt, 333 Cedar Street, New Haven 11, Conn., U.S.A. They should be typewritten in double space, with tabular matter and passages for small print on separate sheets. All materials should be mailed flat.

REFERENCES should be arranged alphabetically and cited by superior numerals in the text. The following order should be used:

Articles: Name of author, full title of article, name of periodical, year, volume, first and last page of article. Abbreviations of names of periodicals should conform to the style used in *A world list of scientific periodicals published in the years 1900–1933* (Oxford University Press, 1934).

Books: Name of author, full title of book, city, publisher, year, edition, full pagination (if book is in more than one volume, give the number of volumes).

ILLUSTRATIONS. Care should be taken not to use too many illustrations, since the Journal provides no figures free of charge. Color plates also are inserted at the expense of the contributor. If illustrations are larger than 8.5×11 inches, reduced glossy prints should be submitted instead.

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